A RETROSPECTIVE OBSERVATIONAL STUDY TO ASSESS THE POST-OPERATIVE FUNCTIONAL OUTCOMES FOLLOWING MODIFIED WOODWARD'S PROCEDURE IN PATIENTS WITH SPRENGEL'S DEFORMITY

A DISSERTATION SUBMITTED IN PARTIAL FULFILLMENT OF THE REQUIREMENT OF **THE TAMILNADU DR.MGR MEDICAL UNIVERSITY**, CHENNAI, TAMILNADU, FOR THE DEGREE OF **M.S. ORTHOPAEDICS** TO BE HELD IN APRIL 2016

A retrospective observational study to assess the post-operative functional outcomes following Modified Woodward's procedure in patients with Sprengel's deformity

Submitted to

THE TAMIL NADU DR. M.G.R. MEDICAL UNIVERSITY

in partial fulfilment of the requirement

for the award of the degree of

MASTER OF SURGERY

in

ORTHOPAEDICS

by

ANAND. A

REG NO: 221312402.MS

THE DECLARATION

This is to certify that the dissertation entitled ″A retrospective observational study to assess the post-operative functional outcomes following Modified Woodward's procedure in patients with Sprengel's deformity" is a bonafide work done by Dr. Anand. A, Christian Medical College, Vellore in partial fulfillment of the University rules and regulations towards MS-ORTHOPAEDICS examination to be held in April 2016 under my guidance and supervision during the academic year 2013-2016.

Candidate:	**Guide:**
Dr. Anand,	Dr. VrishaMadhuri,
PG candidate,	Professor & HOD ,
MS Orthopaedics,	Department of Paediatric
CMCH, Vellore-4	Orthopaedics, CMCH,
	Vellore-4

ENDORSEMENT BY HEAD OF THE DEPARTMENT AND PRINCIPAL OF THE INSTITUITION

THIS IS TO CERTIFY THAT THIS DISSERTATION ENTITLED 'A RETROSPECTIVE OBSERVATIONAL STUDY TO ASSESS THE POST-OPERATIVE FUNCTIONAL OUTCOMES FOLLOWING MODIFIED WOODWARD'S PROCEDURE IN PATIENT'S WITH SPRENGELS'S DEFORMITY' IS A BONAFIDE RESEARCH WORK DONE BY **DR.ANAND.A**, UNDER THE GUIDANCE OF **PROF. DR.VRISHA MADHURI**, PAEDIATRIC ORTHOPAEDIC UNIT, DEPARTMENT OF ORTHOPAEDICS, CHRISTIAN MEDICAL COLLEGE, VELLORE.

DR.V.T.K.TITUS

PROF AND HEAD,

DEPARTMENT OF ORTHOPAEDICS,

CHRISTIAN MEDICAL COLLEGE,

TAMIL NADU, INDIA.

DR.ALFRED JOB DANIEL

PROF AND PRINCIPAL,

CHRISTIAN MEDICAL COLLEGE,

TAMILNADU, INDIA

CERTIFICATE

THIS IS TO CERTIFY THAT THE DISSERTATION ENTITLED ‘‘A RETROSPECTIVE OBSERVATIONAL STUDY TO ASSESS THE POST-OPERATIVE FUNCTIONAL OUTCOMES FOLLOWING MODIFIED WOODWARD’S PROCEDURE IN PATIENTS WITH SPRENGELS’S DEFORMITY’ IS A BONAFIDE RESEARCH WORK DONE BY **DR.ANAND.A**, IN THE PARTIAL FULFILLMENT OF THE REQUIREMENT FOR THE **M.S. DEGREE (ORTHOPAEDICS)** OF THE TAMILNADU DR.M.G.R. MEDICAL UNIVERSITY, CHENNAI TO BE HELD IN APRIL 2016.

GUIDE:

Dr.VRISHA MADHURI
Professor,
Department of Paediatirc Orthopaedics,
Christian Medical College Hospital,
Vellore.

DECLARATION CERTIFICATE

This is to certify that the dissertation titled 'A retrospective observational study to assess the post-operative functional outcomes following Modified Woodward's procedure in patients with Sprengel's deformity' which is submitted by me in partial fulfillment of the requirement of The Tamilnadu Dr. MGR Medical University, Chennai, Tamilnadu, for the degree of M.S. Orthopaedics to be held in April 2016 comprises my original work and due acknowledgement has been made in the text for all material used.

Signature:

Dr.*ANAND.A.*

PG Registrar, Department of Orthopaedics

Chrisitian Medical College, Vellore.

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Lastly but most importantly parents and my sister for their countless love, support and prayers throughout my MS course, this dissertation and forever.

TURNITIN DIGITAL RECEIPT

TURNITIN CERTIFICATE

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Additional Vice Principal (Research)

March 05, 2015

Dr. Anand. A
PG Registrar
Department of Orthopaedics
Christian Medical College, Vellore 632 004

Sub: **Fluid Research Grant Project:**
A retrospective observational study to assess the postoperative functional outcomes following modified Woodward's procedure in patients with Sprengel's deformity.
Dr. Anand. A, PG Registrar, Orthopaedics, Dr. Virsha Madhuri, Orthopaedics, Dr. Thomas Palocren, Orthopaedics, Dr. Abhay Gahukamble, Paediatric Orthopaedics, CMC, Vellore.

Ref: IRB Min No: 9266 [OBSERVE] dated 12.01.2015

Dear Dr. Anand. A,

The Institutional Review Board (Blue, Research and Ethics Committee) of the Christian Medical College, Vellore, reviewed and discussed your project entitled "A retrospective observational study to assess the postoperative functional outcomes following modified Woodward's procedure in patients with Sprengel's deformity." on January 12th 2015.

The Committees reviewed the following documents:

1. IRB Application format
2. Curriculum Vitae' of Drs. Anand. A, Virsha Madhuri, Thomas Palocren, Abhay Gahukamble
3. Informed consent form, Information Sheet & Assent form
4. No of documents 1 – 4

The following Institutional Review Board (Blue, Research & Ethics Committee) members were present at the meeting held on January 12th 2015 in the CREST/SACN Conference Room, Christian Medical College, Bagayam, Vellore 632002.

Ethics Committee Blue, Office of Research, 1st Floor, Carman Block, Christian Medical College, Vellore, Tamil Nadu 632 002.
Tel : 0416 - 2284294, 2284202 Fax : 0416 - 2262788, 2284481 E-mail : research@cmcvellore.ac.in

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Deputy Chairperson
Secretary, Ethics Committee, IRB
Additional Vice Principal (Research)

Name	Qualification	Designation	Other Affiliations
Dr. Inian Samarasam	MS, FRCS, FRACS	Professor, Surgery, CMC	Internal, Clinician
Dr. Anand Zachariah	MBBS, PhD	Professor, Medicine, CMC	Internal, Clinician
Dr. Mathew Joseph	MBBS, MCH	Professor, Neurosurgery, CMC	Internal, Clinician
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Dr. Anup Ramachandran	Ph. D	The Wellcome Trust Research Laboratory Gastrointestinal Sciences, CMC	Internal, Basic Medical Scientist
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Dr. T. Balamugesh	MBBS, MD(Int Med), DM, FCCP (USA)	Professor, Pulmonary Medicine, CMC	Internal, Clinician
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Dr. Anuradha Rose	MBBS, MD	Assistant Professor, Community Health	Internal, Clinician

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Ethics Committee Blue, Office of Research, 1st Floor, Carman Block, Christian Medical College, Vellore, Tamil Nadu 632 002.
Tel : 0416 - 2284294, 2284202 Fax : 0416 - 2262788, 2284481 E-mail : research@cmcvellore.ac.in

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INSTITUTIONAL REVIEW BOARD (IRB)
CHRISTIAN MEDICAL COLLEGE, VELLORE, INDIA.

Dr. B.J. Prashantham, M.A., M.A., Dr. Min (Clinical)
Director, Christian Counseling Center,
Chairperson, Ethics Committee.

Dr. Alfred Job Daniel, D Ortho, MS Ortho, DNB Ortho
Chairperson, Research Committee & Principal

Dr. Nihal Thomas,
MD., MNAMS., DNB (Endo), FRACP (Endo), FRCP (Edin), FRCP (Glasg)
Deputy Chairperson
Secretary, Ethics Committee, IRB
Additional Vice Principal (Research)

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We approve the project to be conducted as presented.

The Institutional Ethics Committee expects to be informed about the progress of the project, any **adverse events** occurring in the course of the project, any **amendments in the protocol and the patient information / informed consent**. On completion of the study you are expected to submit a copy of the **final report**. Respective forms can be downloaded from the following link: http://172.16.11.136/Research/IRB_Polices.html in the CMC Intranet and in the CMC website link address: http://www.cmch-vellore.edu/static/research/Index.html.

Fluid Grant Allocation:

A sum of 15,870/- INR (Rupees Fifteen Thousand Eight Hundred and Seventy only) will be granted for 2 years.

Yours sincerely

B.J. Prashantham

Dr. B.J. Prashantham
Chairperson (Ethics Committee)
Institutional Review Board

Cc: Dr. Virsha Madhuri, Orthopaedics, CMC, Vellore.

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Ethics Committee Blue, Office of Research, 1st Floor, Carman Block, Christian Medical College, Vellore, Tamil Nadu 632 002.
Tel : 0416 - 2284294, 2284202 Fax : 0416 - 2262788, 2284481 E-mail : research@cmcvellore.ac.in

ABSTRACT

TITLE: A RETROSPECTIVE OBSERVATIONAL STUDY TO ASSESS THE POST-OPERATIVE FUNCTIONAL OUTCOMES FOLLOWING MODIFIED WOODWARD'S PROCEDURE IN PATIENTS WITH SPRENGELS'S DEFORMITY

AIM:

To evaluate the Cosmetic, radiological and functional outcomes of modified Woodward's procedure in patients with Sprengel's deformity.

OBJECTIVES:

1. To assess Cavendish score for cosmetic improvement
2. To assess Rigault's score, height-width ratio of the scapula and superior displacement ratio for radiological improvement.
3. To assess the PODCI (Paediatric outcomes data collection instrument)

and simple shoulder test (SST) shoulder scores for functional improvement

METHODS:

In this retrospective observational study, fourteen out of twenty two patients with Sprengel's deformity who underwent modified Woodward's procedure with a minimum of one year follow-up were included and examined. The patients who underwent surgery between January 2006 – August, 2014 under the Paediatric Orthopaedics unit in CMCH Vellore were included in this study. Preoperative findings were noted including patient's detailed history and clinical, radiological & functional evaluation were done at follow-up.

Cavendish grading was used to evaluate cosmetic appearance. Range of motion, Paediatric outcomes data collection instrument (PODCI) and simple shoulder test (SST) were obtained to evaluate shoulder function. Scapula placement and degenerative disease were assessed by radiographic examination and by the Rigault's classification.

Statistical analysis was done using STATA v.13 software. Paired t-test was used for the paired analysis of preoperative and final values of Cavendish grade, range of abduction, ratio of superior displacement and height- width ratio. The Rigault grade and Forward flexion are not normally distributed in the population; we therefore used the Wilcoxon signed rank test

RESULTS:

Out of twenty two Children who underwent Modified Woodward's surgery, only fourteen patients were available for follow- up . No patients had prior shoulder surgery. Out of fourteen patients, three were boys, eleven were girls. They were operated at a mean age of 5.7 years (range 3-12). Four patients had been operated on right side, nine patients on left & one underwent bilateral correction. Six patients had associated anomalies. Mean follow up was 54 months with the mean abduction of the shoulder improved from 107.1^0 preoperatively (90-130) to 143.57 post-operatively ($100\text{-}170^0$). The mean forward flexion* improved from 120.0 preoperatively (100.0,130.0) to 160.0 post-operatively (150.0,160.0). The abduction range of movements had an improvement of 36.5^0 with significant increase ($P<0.001$). The forward flexion also showed a statistically significant improvement of 40^0 ($P=0.0008$). There was a mean cosmetic improvement by Two Cavendish Grades in all 14 shoulders. The preoperative Cavendish grade mean was 3.43 to post-op 1.42 with a significant P-value <0.001.Radiological improvement of scapular lowering was assessed by Rigualt's grade* which showed preoperatively mean 2.5 (2.0, 3.0) to post-op 1.0 (1.0, 1.0) grade with significant p-value of 0.0006. Other radiological assessment such as Superior displacement ratio showed significant improvement with p-value of 0.0001 (pre-op-0.40 compared to post-op of 0.28). Height/width ratio was improved with a p-value of 0.008. No signs of degenerative changes were found in shoulder joints at follow-up radiologically. The mean scapular lowering was 2.01cm. Evaluation of questionnaires with PODCI score & SST at follow-up showed mean PODCI Score of 83.21 (Range – 58-100) & mean SST score of 9.7 points. (Range 8-12). One patient who was operated at the age of 5 years sustained a post-operative brachial plexus inury , however she was not included in the study due to inadequate follow-up. No patients had surgical scar complications like wound necrosis or keloid formation. Hypertrophic scar formation was seen in two patients.

CONCLUSION:

In conclusion our study showed significant improvement in cosmesis, radiological parameters, range of motion, scapular position and function following the Modified Woodward's procedure. The results obtained were in concordance with the published literature. We find this a safe and predictable method with very few complications. We suggest avoiding the modification of anchoring the inferior angle of scapula to the T9 spinous process based on the single significant complication of a brachial plexus injury in a 5 years old child.

Contents

INTRODUCTION

Sprengel's deformity is a congenital elevation of the scapula due to failure of descent during embryonic development. It is a rare anomaly characterized by one shoulder blade placed higher than the other. This complex anomaly is associated with scapular dysplasia and muscular hypoplasia around the region of the scapula. Though rare, it is the commonest anomaly of the shoulder girdle. Though both shoulders can be affected, the left shoulder seems to be affected more often. Among the sexes, it is more common among females with the female: male ratio being about 3:1. It is usually associated with conditions like Klippel Feil anomaly (approximately 1/3 have Sprengel's deformity), scoliosis, omovertebral bar, spina bifida, torticollis, and clavicular abnormalities.

The dysplastic scapula is located higher than normal in the neck or upper thoracic region. This bone is smaller than normal in the vertical plane and appears larger horizontally. The inferior angle is rotated medially and proximally causing the glenoid to face inferiorly.

Limited shoulder abduction and cosmesis are the major concerns. In this condition there are a number of procedures described to correct the above deformity. Of the procedures described the modified Woodward's procedure has been found to be particularly beneficial. However, very few studies describe the functional outcome of surgical correction of Sprengel's deformity. In this study we have assessed the outcome of modified Woodward's procedure which consists of detachment of the origins of the trapezius, division of rhomboid & levator scapulae muscles from the spinous process, caudal displacement of the elevated scapula after excision of any omovertebral bone or fibrous connection on the scapula and excision of the superomedial corner of the scapula. The scapula and detached muscles are relocated at a lower level with heavy sutures and are gradually rehabilitated postoperatively using slings and exercises.

The purpose of this study was to analyse the cosmetic appearance, radiographic improvement & functional outcome of the shoulder at follow-up in a group of patients with Sprengel's deformity who underwent modified Woodward's procedure, the primary procedure in use at our institution. Cosmesis was assessed with Cavendish score and radiological parameter's by Rigault's score pre and post operatively to assess the severity of the deformity, the position of the scapula relative to the cervical spine before and after surgery. The radiographic assessment also included measurement of the superior displacement ratio and height-to-width ratio of the scapula in the posterior scapular view from preoperative images such as chest radiographs and compared with post-operative images. Functional and subjective evaluation have also been assessed using the PODCI (paediatric outcomes data collection instrument) and simple shoulder test (SST).

AIM

To evaluate the cosmetic, radiological and functional outcomes of Modified Woodward's procedure in patients with Sprengel's deformity

OBJECTIVES

1. To assess Cavendish score for cosmetic improvement
2. To assess Rigault's score, height-width ratio of the scapula and superior displacement ratio for radiological improvement.
3. To access the PODCI (Paediatric outcomes data collection instrument) and simple shoulder test (SST) shoulder scores for functional improvement

LITERATURE REVIEW

EMBRYOLOGY OF SCAPULA

Congenital elevation of the scapula also known as Sprengel’s deformity is the most common congenital abnormality of the shoulder girdle (1) (2). The pathophysiology of Sprengel’s deformity is related in particular to the embryology of the upper extremity.

The formation of scapula occurs during the embryonic period through a process of germ layer differentiation between the third and eighth weeks of pregnancy. The scapula lies at the level of the fourth and fifth cervical vertebrae at this time(2). Continuous cellular signaling from surrounding tissues is required for differentiation of the mesoderm into the axial and appendicular skeleton. The programmed mesenchymal cell pathway is needed for the development of the scapula. Various cell-signaling molecules such as bone morphogenetic proteins and fibroblast growth factors help the pluripotent mesenchymal cells to differentiate into skeletal tissues(3). A group of specialized mesenchymal cells, the apical ectodermal ridge which is present in the periphery of the limb bud, directs underlying limb outgrowth. The scapular growth and development is guided by multifaceted cellular signaling pathways, which in turn direct the growth and development of surrounding muscles, bones, and nerves.

The scapula along with the upper limb develops in the upper dorsal/lower cervical region with the arm bud during the 5^{th} week and descends to 2^{nd} -8^{th} dorsal vertebrae which is the final anatomical position by the 12^{th} weeks of gestation(final location at the level of the seventh thoracic vertebra)(4) (5). The vertical diameter of scapula is more than the horizontal diameter measured at the base of spine of scapula at 12 weeks. The pathology in Sprengel's deformity probably represents a continuation of the fetal form of the scapula. It has been described that the smaller deformed scapula has a vertical diameter smaller than the horizontal diameter, anterior curving of the supra-spinous portion with prolongation of the superior medial scapular angle(6)(7). The anterior curving of the supra-spinous portion with prolongation of the superior medial scapular angle is due to the smaller deformed scapula which has a horizontal diameter that exceeds the vertical diameter. The scapula generally descends to its thoracic location about the twelfth post gestational week, having formed from paraxial mesoderm at about the level of the fourth or fifth cervical vertebra. The usual post migration location of the scapula is between the levels of the second and eighth posterior ribs. The surrounding structures are also affected which require a normal scapula for development.

Sprengel's deformity occurs due to problems with limb bud formation. The most obvious presenting sign is the elevated scapula which is seen along with the hypoplasia of the scapula and surrounding musculature. The bone formation of scapula is via intramembranous ossification. Pectoralis major, trapezius, rhomboids, levator scapulae, serratus anterior and latissimus dorsi are the muscles acting on the scapula which shows varying degrees of involvement of these muscles, seen in congenital elevation of the scapula(8). Trapezius has been reported to be atrophied in cases of acquired elevation of the scapula (9). The trapezius inserts along the medial border of the scapula and resists the upward-directed forces of the levator scapulae and the rhomboids.

Few proposed theories for the cause of these syndromes are inheritance of an autosomal dominant pattern, vascular lesions arising from the subclavian artery(8)(10). Most cases are sporadic, and the etiology remains unknown. But it is unclear whether inheritance of autosomal or vascular lesions are responsible for Sprengel's deformity, but they may not mutually exclusive. Other patho-anatomical findings include an omo-vertebral bar, articulations with the vertebral column; and these may extend from the supero-medial scapular angle or the upper third of the medial border of the scapula up to the transverse process of a cervical vertebra (one of the fourth-to-seventh vertebrae).

Additionally, various other types of disruptions of limb bud formation and differentiation can be noted, including hypoplastic thumb, clavicular hypoplasia, and hemimelias, costo-vertebral defects (spina bifida and kyphoscoliosis) and underdevelopment of pectoral girdle bones (clavicle, humerus) and musculature (pectoralis major, trapezius) may coexist. The other various abnormalities during embryonic phase are Poland anomaly, Klippel-Feil syndrome, and Möbius syndrome (3). Because of the cooperative growth of the skeletal system, such conditions lead to multiple phenotypic abnormalities.

ANATOMY OF THE SCAPULA

The Scapula is a flat bone which lies on the posterolateral aspect of the chest wall which is large and triangular in shape and has two surfaces, three borders, three processes and three angles.

The long axis of Scapula is nearly vertical and it extends from the second to the seventh ribs covering its parts. There are costal and dorsal surfaces; medial, lateral and superior borders and superior, inferior and lateral angles. The head of humerus is in articulation with the glenoid cavity which is borne by the lateral angle which is considered to be the head of the scapula which is connected to the body by an inconspicuous neck.

The scapula has three projections namely, spinous process, acromion and coracoid processes. The lateral border of the scapula is usually thick and extends from the glenoid cavity above to the inferior angle below. The shelf like projection of the spine interrupts the two surfaces of scapula namely the costal and dorsal surface. The free surface of the lateral angle helps in gleno-humeral articulation by bearing the glenoid cavity which is pear shaped, wider below and narrow above (11).

The lateral angle is broad, truncated and constitutes the head of the scapula. The supraglenoid tubercle is a roughened, small area above the glenoid cavity which encroaches on the coracoid process. When the arm is raised above the head the inferior angle of scapula can be seen as it passes forwards around the chest wall, and can be felt through the skin and muscles which cover it. The Inferior and dorsal aspects help in identifying the neck of scapula easily which is a constriction immediately adjoining the head.

The neck of the scapula extends between the infraglenoid tubercle and anterior margin of the suprascapular notch on the ventral aspect(11). Trabecular bone and compact bone form the thicker and thinner parts of the scapula respectively. Fibrous tissue fills up the gap in the bone which is deficient over the central supraspinous fossa and the greater part of infraspinous fossa which are usually thin and translucent.

Dorsal Surface:

The shelf like spine of the scapula divides the dorsal surface into an upper part which is small and lower part which is large. The dorsal surface is confluent at the spino-glenoid notch between the dorsal aspect of neck and lateral border of spine. The muscles which are attached on the dorsal surfaces are the Supraspinatus, Infraspinatus and the Teres minor. The medial two-thirds of the supra-spinous fossa is the attachment of the Supraspinatus. The fascia which covers this muscle is attached to the margins of the fossa. The Infraspinatus is attached to the infraspinous fossa sparing an area near the neck of the bone. Teres minor is attached to the upper two thirds of flattened strip on the dorsal surface which continues with the lateral border. Attachment of the lower limit of the Teres minor is identified by an oblique ridge (runs from lateral border to inferior angle) and cuts off an oval area where the Teres major is attached. The circumflex scapular vessels which forms a groove near the upper end of the strip enters the infraspinous fossa after passing between the Teres minor and bone. The inferior angle on the dorsal aspect gives origin to a small slip which joins the deep surface of Lattismus dorsi. The infraspinatous fascia which partitions the teres major and teres minor mark the limit of their attachment.

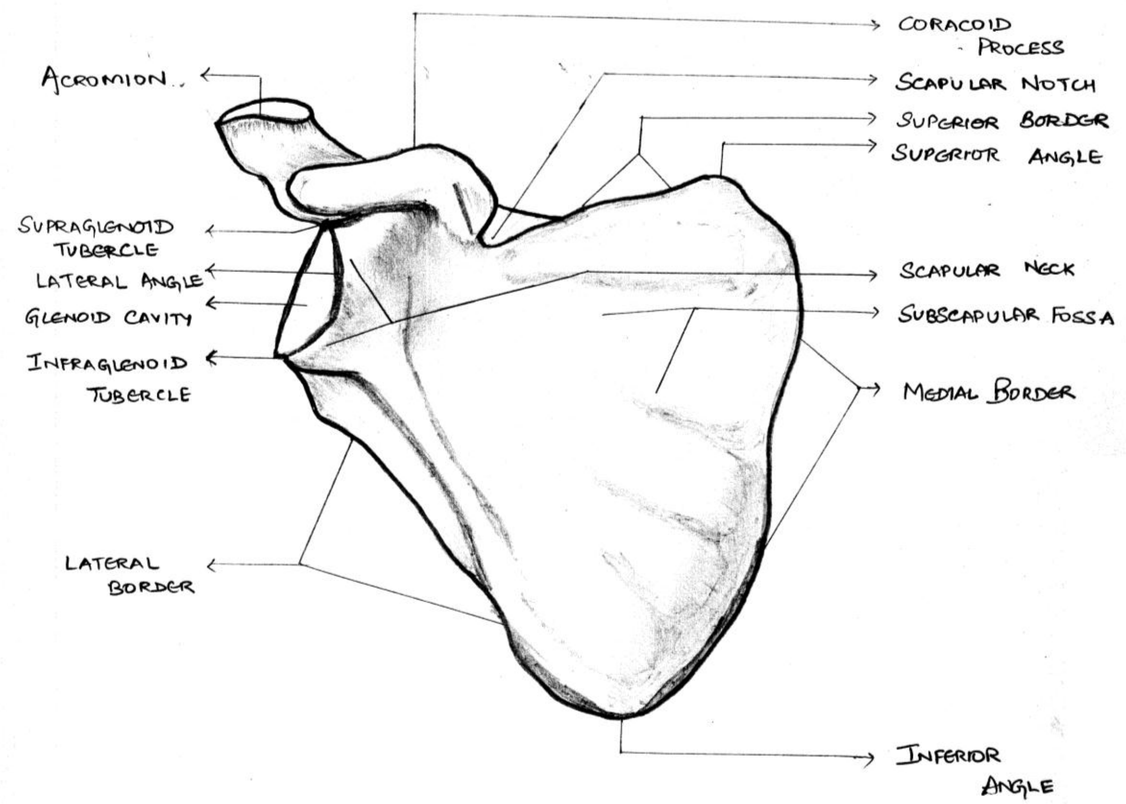

Figure 1.Scapula anatomy - anterior view.

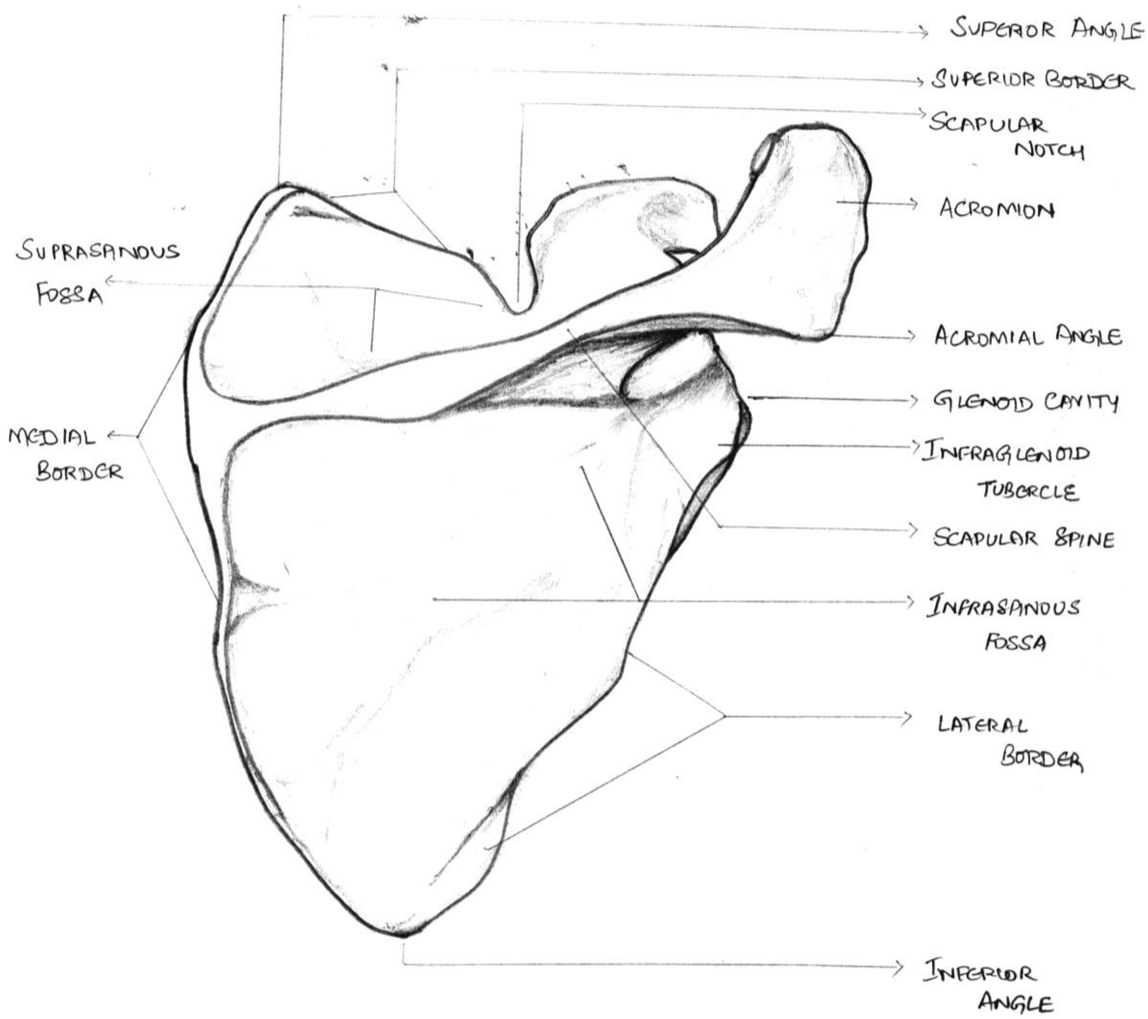

Figure 2. Scapula - posterior view

Scapular Angles:

The superior angle is obscured by the upper part of the trapezius at the junction of superior and medial borders. The lateral angle helps in gleno-humeral articulation by bearing the glenoid cavity which is pear shaped, wider below and narrow above. The lateral angle is broad, and truncated. When the arm is raised above the head the inferior angle of scapula is visible as it passes round the thoracic wall, which is also palpable through the skin and muscles which cover it. The upper border of the Lattismus dorsi covers the dorsal aspect by a small slip from which it is frequently attached to the inferior angle. The most distinct feature at the dorsal and inferior aspects are the anatomical neck and the constriction adjoining the rim of the glenoid cavity. Anteriorly and posteriorly it extends between the Infraglenoid and Supraglenoid tubercles. The Supraglenoid tubercle serves as an attachment for the long head of the Biceps and the Infraglenoid tubercle for the long head of the Triceps (11).

Superior Border:

The suprascapular notch separates the anterolateral end of the superior border from the coracoid process. The inferior belly of the Omohyoid originates near the suprascapular notch. The superior transverse ligament bridges the suprascapular notch, which is attached medially to the limit of the notch and laterally to the root of the coracoid process(11). The superior border is the shortest of all three borders, sharp and thin. The suprascapular vessels pass backwards above the ligament while the foramen transmits the suprascapular nerve to the supraspinous fossa.

Medial Border:

Medial border is thin, most often angled opposite to the root of the spine , extends from the superior to the inferior angle. The medial border is easily felt in the lower two thirds while it cannot be palpated in the upper third as it is more deeply placed. The muscles attached to the upper part are the Rhomboid major, Rhomboid minor and the Levator Scapulae. Levator scapulae extends from superior angle to root of spine and attached to a narrow strip whereas the Rhomboids minor is attached below this opposite to the root of spine and Rhomboids major to the remainder of the border.

Lateral Border:

It forms a sharp, well defined roughened ridge which runs continuously from the inferior angle of the scapula to the glenoid cavity. The Infraglenoid tubercle is a triangular area on upper end of the lateral border. The infraglenoid tubercle serves as an attachment for the long head of triceps. The lateral border separates the attachment of subscapularis, Teres minor and major (11). The lateral border of the spine cannot be felt through the skin as the Lattismus dorsi muscle covers it completely.

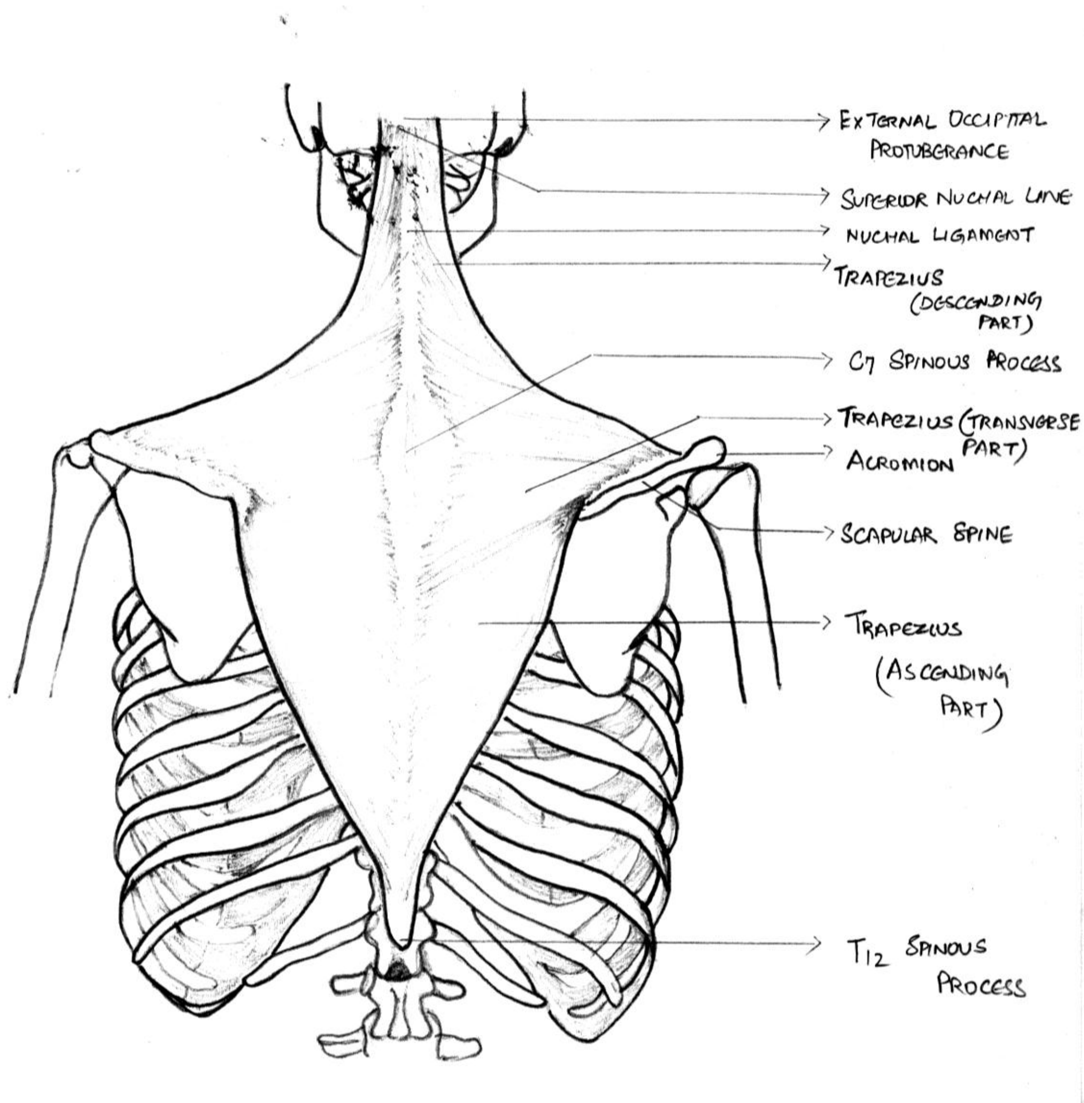

Figure 3. Trapezius - posterior view.

Spine of The Scapula:

The spine of the scapula is triangular in shape, forms a shelf like projection on the upper part of the dorsal surface of the bone. The anterior border of the spine of the scapula joins the dorsal surface of the scapula along a line which runs slightly upwards and laterally from the junction of the upper and middle third of the medial border(11). Concavity of the upper part of the costal surface is formed as the plate like body of bone is bent along this line. The crest of the spine forms the dorsal border and it is subcutaneous throughout nearly its whole extent. It expands into a smooth, triangular area on the medial end. The remaining parts of the surface of crest along with the upper and lower edges are roughened for muscular attachment. The spinoglenoid notch lies between the lateral border and the dorsal surface of the neck of the scapula. The Infraspinatus and Supraspinatus are attached to the lower and upper surfaces of the spine of the scapula.

The flattened triangular area at its root lies opposite the spine of the third thoracic vertebra and is covered by the tendon of the trapezius. The upper border of the crest serves as an attachment for the middle fibres of the trapezius, whereas the lowest fibres of the trapezius terminate in a flat triangular tendon which glides over the smooth area at the base of spine and inserts into the deltoid tubercle on the dorsal or the medial end near the subcutaneous aspect of spine(11).

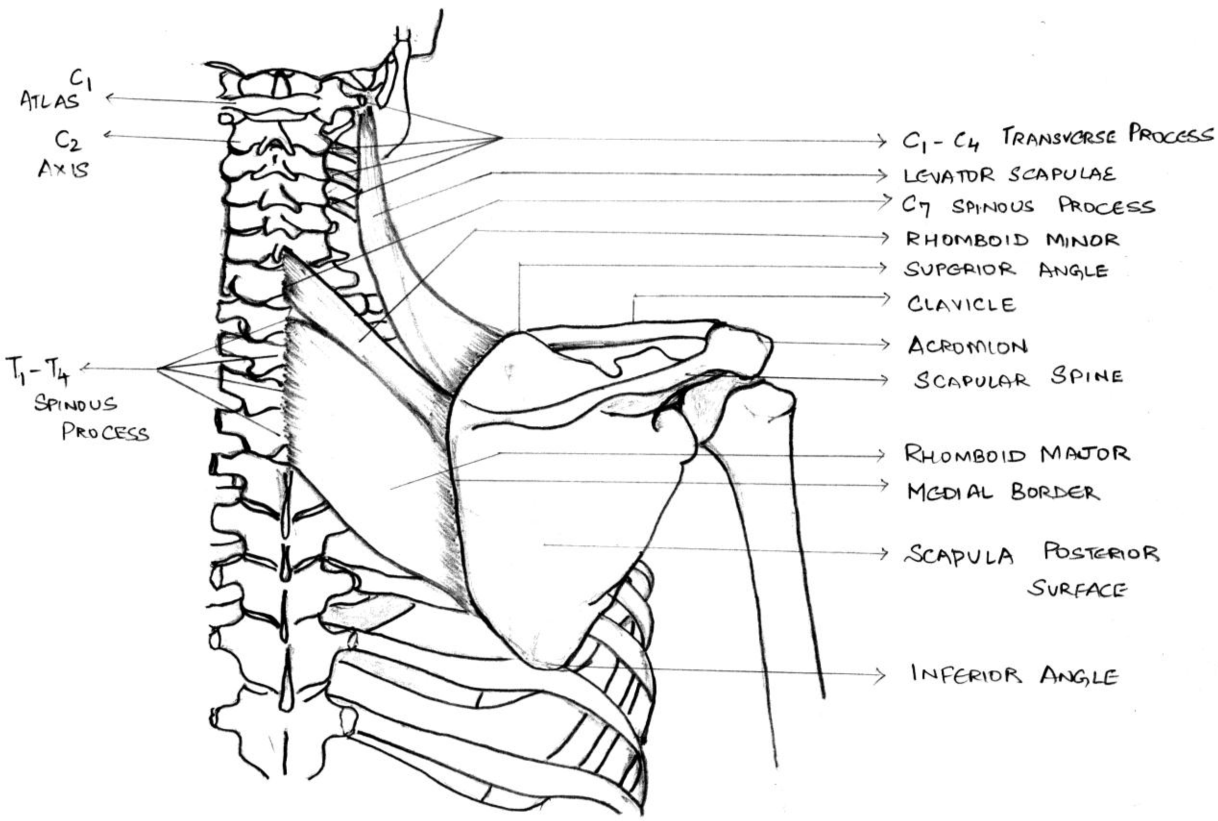

Figure 4.Levator scapulae with Rhomboids major and minor.

Scapulothoracic interface:

Scapulothoracic movements are of a gliding nature and occur at an interface between the ventral surface of the scapula and the rib cage. The contacting surfaces involve the subscapularis and bare areas of the scapula with the serratus anterior overlying the second through the seventh ribs. Although the scapula has no bony or ligamentous connections with the thorax, numerous muscles hold the scapula in close opposition to the chest wall like the serratus anterior, trapezius and rhomboids. Weakness or paralysis of these same three muscles results in winging of the scapula. With the upper limb in a neutral position and at rest, inspection can lead to the anatomic diagnosis of scapular winging. Weakness of the rhomboids results in a resting position where the scapula is directed laterally (protracted) and rotated upward (As a result of the antagonist muscles pulling without opposition). Likewise, weakness of the serratus anterior results in a retracted scapula (directed medially) and rotated downward. Weakness of the trapezius results in a protracted scapula (directed laterally) and rotated downwards. Normally the scapula is set obliquely on the thorax at an angle of 30, open anterolaterally, and moves along a curved thoracic surface during protraction and retraction

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SPRENGEL'S DEFORMITY

Sprengel's deformity is most often seen in children, as a congenital anomaly (12) in which the children have a scapula that sits too high mostly on one side. Normally during fetal development, the scapula moves down the back to rest in its normal position. These children will have cosmetic problems, limitations of movement especially abduction and forward flexion of the affected shoulder. Associated anomalies like Klippel-Feil syndrome and scoliosis often co-exist and affected children are at greater risk of developing renal disease.

DEFINITION

It is defined as a rare congenital anomaly. During embryological development which causes failure of normal descent of the scapula, from its position in the neck, to its normal position in the posterior thorax. The affected scapula is smaller and more cephalad than the normal scapula which is characterised by medial rotation of the inferior part of the affected scapula and elevation(6) (13). The limitation of shoulder movement and disfigurement is caused by regional muscle hypoplasia or atrophy. The deformity is usually unilateral, but it can be bilateral also(5)(13)(15). Sprengel's deformity can be isolated or part of a syndrome. In around a third of patients, the affected scapula is attached to the cervical spine by an omovertebral bone(cartilage or fibrous tissue), which when present, makes abduction of shoulder beyond 90 degrees mostly impossible(16)(7)(17).

Sprengel's deformity is characterized by a high-riding scapula, asymmetry in the shoulder contour and restriction of shoulder movement. It is caused by a variable arrest in the descent of the scapula during intrauterine development.

Michel Mortiz Eulenberg described the deformity as high grade dislocation of scapula (hochgradige dislocation der scapula) in 1863 in three patients(18). After about 22 years in 1863, Willet and Walsham were the first to describe the omovertebral bone and gave a comprehensive anatomical description, the methods of its excision and reported good results of it. Sprengel's deformity is associated with a variety of other congenital anomalies. It was Sprengel in 1891, who illustrated this deformity in four cases and hence its name. Howitz in 1908, Fairank in 1914 and Grieg in 1924 reported few cases.

The exact cause of this congenital deformity or presence of the omovertebral bone is still unknown, but it is clear that during the embryonic phase a disruption of the migration of the scapula occurs from the embryonic limb bud level opposite C5, which also causes failure of normal bone and soft tissue development in the shoulder girdle (19) (20) (21). The hypothesis is that all these tissues and the scapula are of mesodermal origin and develop in the same embryonic period. Failure can therefore result in several associated anomalies (22). Depending on the severity, the deformity could be obvious at birth or manifest later in childhood.

Occasionally Sprengel's deformity could also occur as part of the Klippel-Feil Syndrome (in 30% cases) or could be associated with other spinal and cranial anomalies or absent ribs(6) (13) (23). The superior part of the scapula may be prominent and curved forwards over the apex of the thorax leading to deformity of the clavicle. Shoulder musculature is hypoplastic, fibrotic, or even absent in most cases, and weakness of the serratus anterior muscle may cause scapular winging (24) (16).

In 20-30% of the patients, an omovertebral ‘‘bone’’ is found, either consisting of bone, cartilage, or fibrous tissue, between the superior angle of the scapula and the apophysis of the spinous process of the cervical vertebrae.

EPIDEMIOLOGY

Sprengel’s deformity is the most common congenital malformation of the shoulder girdle. In general, there is predominance for the female gender [3:1] (19) (20) and usually one shoulder is affected; but bilateral involvement has been described(7) (25). This often painless condition causes partial impairment of the shoulder movement, especially abduction and scapular-thoracic rotation, causing functional disability and disruption of normal cosmetic appearance. It is unknown if this altered anatomical situation leads to early degenerative changes in the shoulder joint. The main goal of surgery for Sprengel’s deformity is to improve shoulder function as well as the cosmetic appearance.

AETIOLOGY

The condition is sporadic. Rarely, it shows an autosomal dominant pattern of inheritance, which is known as Corno's disease (26) (27).

CLINICAL FEATURES

Usually children present with complaints of affected scapula being located higher than normal, by around 2 to 10 centimeters. The scapula appears to be larger than normal horizontally and is adducted. It also appears smaller than normal in the vertical plane. The glenoid appears to be facing inferiorly since the inferior angle is medially rotated. A characteristic visible lump is seen in suprascapular region due to the superomedial angle of the scapula which is upwardly rotated, causing the ipsilateral side of the neck to appear fuller. Lower the scapula is rotated more medially.

Usually an omovertebral connection exists which is trapezoid or rhomboid shaped in structure. An omovertebral connection exists in about one third of cases (16) (17). This is a rhomboid or trapezoid shaped structure that usually lies in a strong fascial sheath, which extends from the superomedial angle of the scapula to the spinous process, lamina or transverse process of the cervical vertebrae (most commonly the fourth to seventh cervical vertebrae). It may be fibrous, cartilaginous or bony. The omovertebral connection is usually unilateral. It is the primary cause of restricted shoulder motion in patients with Sprengel's deformity. It is always associated with a fixed, elevated scapula and has a major role in determining the shape and the malpositioning of the scapula. The spinoscapular muscles are also adversely affected. Trapezius, levator scapulae, pectoralis major, rhomboid, serratus anterior, latissimus dorsi or the sternocleidomastoid muscle may be absent, hypoplastic or contain multiple fibrous adhesions. The trapezius muscle is the most commonly affected muscle. Winging of the scapula may occur if the serratus anterior muscle is weak. Usually Sprengel's deformity is painless. On examination, passive movement of the glenohumeral joint, including initial abduction, internal and external rotation may be normal. Usually the scapulothoracic movements are limited. The most common limitations are abduction and forward flexion. Abduction is limited to less than one hundred degree in 40% of patients which is more likely if an omovertebral bone is present.

Clinically the scapular fixation causes restriction of shoulder elevation and this is due to three factors

- the omovertebral bone
- the medial border abutting against the spinous process of adjacent vertebrae
- the superior angle of scapula being curved forwards over the apex of the thorax

ASSOCIATED ABNORMALITIES

Sprengel's deformity mostly never occurs as an isolated malformation. It is usually accompanied by various other anomalies, especially in the cervico-thoracic vertebrae or the thoracic rib cage. The most common associated defect is Scoliosis, Klippel-Feil's syndrome or fused ribs, chest wall asymmetry, congenital cervical spina bifida, or cervical ribs. Most common anomalies were hypoplasia or absence of pectoralis major, rhomboids, trapezius, lattismusdorsi and serattus anterior. Eulenberg and Greig(1911) considered shortening of the levator scapulae as an important etiological factor as it was probably present in most cases of any severity.

Syndromes associated with this condition include Greig syndrome (28) (characterised by polydactyly, cutaneous syndactyly, ocular hypertelorism, macrocephaly and a high, prominent forehead), Poland syndrome (29) (characterised by hypoplasia or absence of the pectoralis on one side of the body & cutaneous syndactyly of the ipsilateral hand), Klippel-Feil syndrome and VATER association (30) (characterised by vertebral defects, imperforate anus, tracheoesophageal fistula, radial dysplasia and renal dysplasia). The most common syndrome is Klippel-Feil syndrome. Other rarer syndromes include DiGeorge Syndrome (31) (characterised by heart defects, cleft palate, autism, learning disabilities, recurrent infections and hypocalcemia) ,Floating-harbor syndrome (characterised by short stature, delayed bone growth, delayed communication skills and distinct facial features), Goldenhar syndrome (characterised by incomplete development of the ear, nose, soft palate, lip, and mandible). X-linked dominant hydrocephalus and mental disturbance syndrome. A relationship between Diastematomyelia (a condition in which a part of the spinal cord is split, usually at the level of the upper lumbar vertebrae) and Sprengel's deformity has also been described.

CLASSIFICATION

Cavendish Grading is widely used and recommended for grading cosmesis in Sprengel's deformity. Grade 1 is the mildest, where the shoulders are almost level and it cannot be noticed with clothes on. Grade 2 is also mild, but the superomedial portion of the scapula is visible as a lump. In Grade 3, the deformity is moderate, visible and the affected shoulder is two to five centimetres higher than the normal shoulder. In Grade 4, the deformity is severe, the scapula is very high, with the superomedial angle at the occiput, with neck webbing and brevicollis. One limitation of this classification is that it is difficult to apply in bilateral cases.

IMAGING

Postero-anterior radiograph of the chest and both shoulders is the best way to identify Sprengel's deformity. Leibovic et al described a method to calculate the scapular displacement ratio, which uses three lines drawn on postero-anterior radiograph, which helps to calculate the superior scapular angle and the inferior scapular angle which give the viewer some idea about scapular rotation (32). CT scans may be performed to visualise the affected region, delineate the attachments of the omovertebral bone or to determine the presence of spina bifida occulta or an intraspinous lesion before surgery (33). Appropriate imaging studies should also be performed for any associated anomalies.

OUTCOME SCORES:

The cosmetic appearance, radiographic improvement& functional outcome of the shoulder at follow-up were assessed in patients with Sprengel's deformity who underwent Modified Woodward's procedure. The Cavendish score and Rigault's radiological score is used to assess the severity of the deformity, the position of the scapula relative to the cervical spine before and after surgery. Functional and subjective evaluation is assessed using the PODCI (paediatric outcomes data collection instrument) and simple shoulder test (SST). Cavendish grading is used to evaluate cosmetic appearance.

CAVENDISH CLASSIFICATION:

Sprengel's deformity patients typically present with cosmetic and functional impairment. In concordance with the current literature, we used the Cavendish classification (16) for objective evaluation of surgical treatment.

This classification is easy to use and evaluate more exactly the amount of deformity and post-surgical improvement. Cavendish in a series of one hundred cases from series of hospital in United Kingdom found moderate to severe deformity in 41 and mild deformity in 59 cases. Errors in diagnosis can occur even though main features of the condition are known. Klippel-Feil syndrome , Erb's palsy and Scoliosis are some of the conditions that causes diagnostic difficulty. It can be differentiated fairly simple if proper clinical and radiological examination is made.

The children affected with Sprengel's deformity have decreased range of motion (ROM) and varied degrees of functional limitation resulting from the shoulder joint.

Lack of motion of the Scapulothoracic junction and an inferiorly rotated glenoid result in decreased shoulder abduction. Shoulder abduction is usually limited to 90°. These limitations affect many activities of daily living. It is not easy to classify a deformity when it is bilateral and deformities are of the same grade. To simplify the indication for treatment and description of results the following grades were formulated by Cavendish in 1972 (16).Grade 1 - Very mild, shoulder joints are level, the deformity is invisible when the patient is dressed. Grade 2 - Mild Lump in the web of the neck and deformity is visible when dressed. Grade -3 there will be elevation of shoulder ~2-5cms and deformity is easily visible. Grade -4 is severe and the shoulder is very elevated such that the superior angle of the scapula is near the occiput ,with or without neck webbing or brevicollis. Cavendish grade is used as a measure of cosmetic improvement and range of abduction at the shoulder as a measure of function(16). Scoliosis was found to be the most common associated abnormality in two large series of 112 patients and 75 patients (35% and 55%, respectively). Rib anomalies are also common, with a reported prevalence of 16% to 48% in patients with Sprengel's deformity. Other associated disorder included diastematomyelia.

Table 1.Cavendish classification.

Cavendish Score	**Sprengel's deformity**
Grade 1 - Very mild.	Shoulder joints are level. Deformity is invisible when patient is dressed.
Grade 2 - Mild.	Lump in the web of the neck. Deformity is visible when dressed.
Grade 3 - Moderate.	Shoulder elevation 2-5 cm. Deformity is easily visible.
Grade 4 - *Severe*	Superior angle of the scapula is near the

	occiput with/without neck webbing or brevicollis

RIGUALT SCORE:

Any patient with Sprengel's deformity requires a radiographic evaluation as initial assessment. It should be obtained initially to assess the level of the scapula in relation to the vertebrae and the contralateral side. It is also helpful to determine the presence of associated abnormalities (eg, scoliosis, rib abnormalities, omovertebral bone).

Rigault's classification described by Rigualt et al is based on the projection of superomedial angle on radiographs(13)(34). It is used to assess scapular level with respect to vertebrae radiographically pre-op and post-operatively. It is simple, easily calculative and widely used scoring. This scoring does not have any correlation with the Cavendish score.

The radiographic assessment included the measurement of the superior displacement ratio and height-to-width ratio of the scapula in the posterior scapular view from preoperative images such as chest radiographs comparison with post-op images. Scapula placement and degenerative disease will be assessed by radiographs. "Scapular Index" suggested by Broca describes the relationship between the vertical length and horizontal breadth of scapula. This index was represented by a formula (100*breadth/length) and calculated to be approximately between 63-71(9).

Plain radiographs can be used for assessing the deformity and presence of omovertebral communication, and to note the postoperative correction. The distances between the inferior and the superomedial angles of the scapula and the spinal column, and the angle of glenoid tilt, to assess the postoperative outcomes was described by Ahmad et al (35) which is not used widely. Based upon the frontal radiographic images and the relation of the superomedial angle to the vertebral column, Rigault proposed a classification to assess the deformity between the superomedial angle of the scapula and the vertebral column.

A radiographic classification for Sprengel's deformity was developed by Rigault et al which is based on radiograph - the projection of the superomedial angle of the scapula. Deformity is graded based on the relationship between the superomedial angle of the scapula and the associated vertebral level. Grade 1 deformity involves the superomedial angle below T_1, grade 2 deformity lies between T_1 and C_5, and grade 3 deformity lies above C_5.

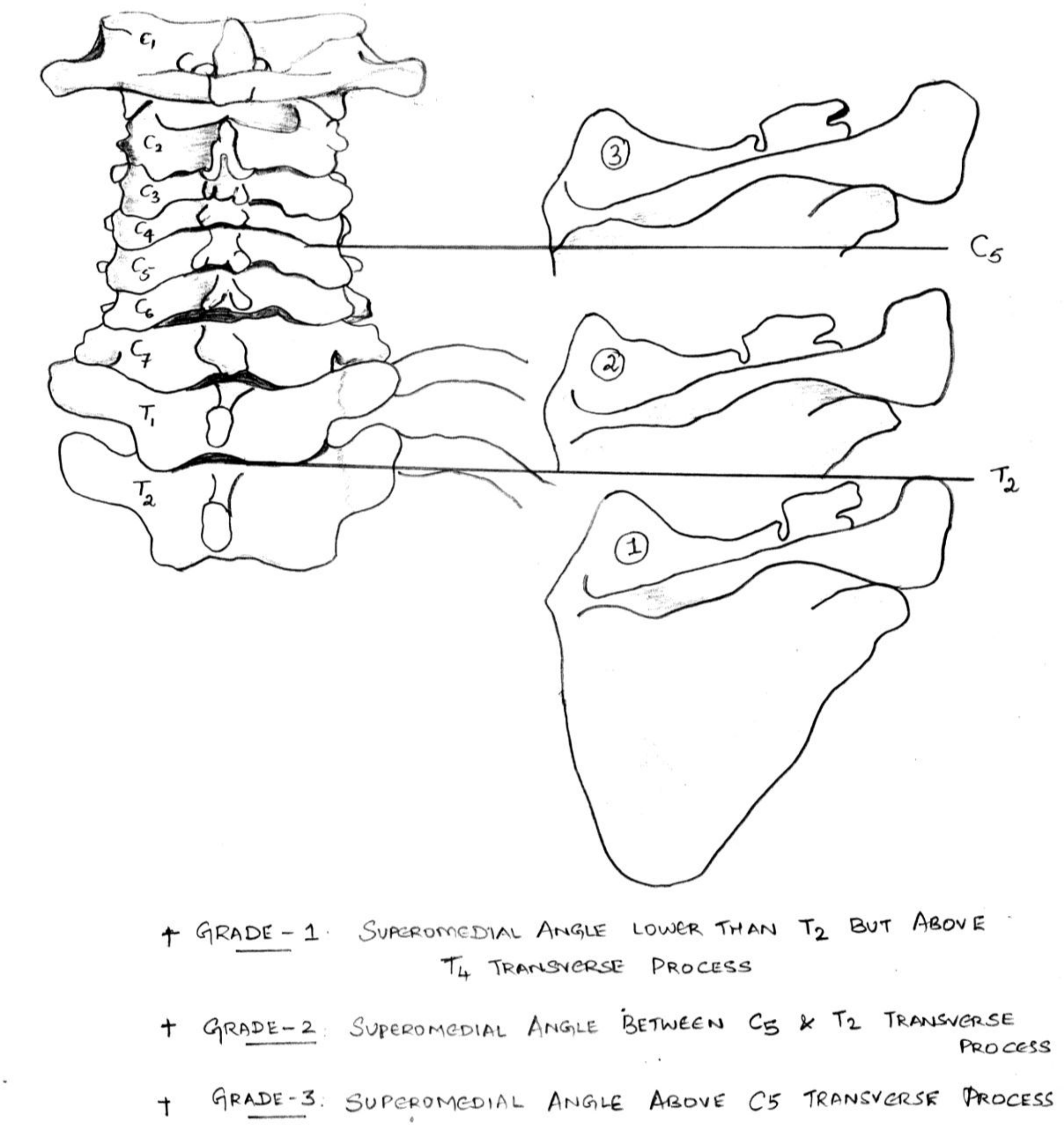


Figure 5. Rigault's radiological clssification of the sprengel's deformity of the shoulder.

Another classification described by Ross and Cruess measured shoulder elevation based on the level of the center of the humeral head in relation to the trunk vertical axis. Leibovic et al described Sprengel’s deformity has a rotational component, and measured scapular displacement based on the vertical positioning of the scapula and the rotational component, assuming that the center of rotation of the scapula was through the acromioclavicular joint(32).

To reveal scapula position, (early) degenerative changes, and anatomical differences between both shoulders standard radiographs of antero-posterior view of both shoulders with chest X-ray was done. In patients who had associated anomalies, measurments of ‘Superior displacement ratio’’ was done by drawing two reference lines. Line 1 will be drawn from the center of the glenoid cavity of the affected shoulder perpendicular to the vertebral axis line, and line 2 was drawn from the normal shoulder. The superior displacement ratio was defined as the distance between these two lines as a fraction of the scapular height on the normal side(33). This ratio is not measured in bilateral cases, as there is no normal side to use as a reference.

The way to assess the height-to-width ratio on the radiograph is the height of the scapula is measured from the superior angle to the inferior angle parallel to the glenoid, and the width of the scapula is measured from the cavity of the glenoid to the medial most portion of the vertebral border perpendicular to the glenoid. To assess scapular lowering after surgery, the inferomedial angle of the scapula was used as the reference point (24) . These measurements were assesed from radiographs pre-operative and post-operatively . Objective measurement of scapular lowering is difficult and controversial due to the hypoplastic nature of the affected scapula. Associated spinal anomalies, scoliosis and hemivertebrae were documented.

Examination of the shoulders and both upper limbs, spines, and lower limbs were done by giving attention to scapular bony prominences, scapular winging, scoliosis, chest asymmetry, or any associated morphological abnormalities. All the patients were examined for the grade of the deformity. The forward flexion and abduction movements of the shoulders were examined, as well as looking for range of motion especially of the scapulo-thoracic motion, to determine whether the scapula is anchored to the spine or not.

PODCI

A number of studies have been done evaluating the severity and outcome of treatment using various Scales like Constant Score, DASH (Disability of Arm,Shoulder and Hand Score). Constant score is not originally meant for assessing children, not standardised for children and the strength component has significant variability and unreliability with varying ages and sex and not a good outcome tool.

PODCI (Pediatric Outcomes Assessment instrument) is designed by the American Academy of Orthopaedic Surgeons (AAOS) along with the Pediatric Orthopaedic Society of North America and the council of musculoskeletal speciality societies, for assessment of individuals with musculoskeletal disorder. It includes children aged 2-19 years taking into account effectiveness of treatment and their quality of life(36). The PODCI subscales were described by Hunsaker et al (37), who described the normative values from the general population which includes upper extremity function(UE), Transfers and basic mobility(TBM), sports and physical function(SPF), comfort, happiness and global function.

The Pediatric and Adolescent Outcomes Instruments are designed to assess patients under 19 years of age with regards to overall health, pain and ability to participate in normal daily activities, as well as in more vigorous activities associated with young people.

According to AAOS and POSNA, development of more generic instruments to compare different disorders and interventions would be effective and beneficial. Knowing the age, sex and comorbidities across conditions thus forms a critical point of assessment in evaluating a child in the current era.

In a study conducted by Lerman et al, PODCI has been used to quantify abilities of patients with unilateral upper extremity deficiencies(U-UED) in comparison with a group of normal children(38). This study included children between 11-21 years. In this study, it was noted that there was a functional difference among the unilateral upper extremity deficiencies patients when compared with the normal control children in PODCI questionnaire. This study suggested that future innovations in prosthetics would overcome the differences between the U-UED and normal control children. There was no significant difference among the congenital and traumatic amputees after controlling the age factor.

In another study conducted by Amor et al, the PODCI was given to parents of 74 children with amyoplasia (39). The score was repeated and the initial score was compared with recent score using student T test. The results of this study suggested that the PODCI is useful in amyoplasia children especially to assess the range of function over time and also useful in the assessment of long term outcomes with surgical management.

Pediatric Outcomes Questionnaire

The Pediatric Outcomes Instrument is designed to be completed by the parent/guardian of a child ten years of age or younger who has knowledge of the child's conditions. This questionnaire is similar to the Adolescent (parent-report) Outcomes Questionnaire, but has slightly different wording and uses different scoring algorithms.

Adolescent (self-report) Outcomes Questionnaire

The Adolescent (self-report) Questionnaire is intended for youth ages 11 to 18 years who are capable of completing the form independently. This questionnaire is similar to the Adolescent (parent-report) Outcomes Questionnaire, but does not offer a response option indicating that the respondent is "too young for this activity."

The individual's Standardized score is based on the mean of items that make up the scale. All standardized scores are calculated in the worksheets such that a 0 represents the MOST disability and 100 represents LEAST disability.

To make the scores comparative across various scales, the Normative Data Study's results were transformed for each scale so that each has a mean Normative Score of 50. Thus, a patient scoring above 50 on a particular scale is above the general population's average, while a patient scoring below 50 on a scale is below the general, healthy population's norm (40).

A mean for the overall scale scores was derived as described earlier and is set at 50, with a standard deviation of 10. Forcing to a set mean and standard deviation rather than using a standard z-score transformation with a mean of 0 and a standard deviation of 1 provides the basis of comparison for the Normative Scoring.

Overall scoring is derived from the functional assessment scores, with each having a possible range from 0-100. Higher scores indicate higher levels of disability and lower scores indicate better functioning for most items.

The PODCI score has been reported to be a valid and reliable instrument.

To Our knowledge PODCI scoring system has never been used before in the evolution of Sprengel’s deformity.

SIMPLE SHOULDER TEST:

A prospective study was conducted by Roy et al in which the patients were evaluated both preoperatively and 6 months post operatively(41). One of the outcome measures used in this study was simple shoulder test (SST). This is a self -questionnaire with 12 yes/ no response questions. The scores were given from 0(worst) to 12(best).

Another study was conducted by Roy et al to evaluate the shoulder function using four different questionnaires. Among the four questionnaires, the minimal detectable change (MDC) and the minimal clinically important difference (CID) have not been defined for SST.

The findings of the prospective study by Roy et al showed that the CID of SST is 3 on the 12 point SST scale. Use of SST as a measure for monitoring the patients at 6 months following shoulder arthroplasty had also been suggested in this study.

SST was found to be quick, reliable and clinically useful. Due to its responsiveness following rotator cuff repair and shoulder arthroplasty, SST has been suggested to be used in places where a brief shoulder specific measure is required by clinicians.

In a study conducted by Walstra E F et al, patients with Sprengel’s deformity corrected with Woodward’s procedure were followed up. Among the various assessment measures used in their follow up, the disabilities faced in work and other everyday activities were evaluated using DASH and SST questionnaires (18). The maximum score of SST was fixed as 13 points. Better shoulder function had higher values. The mean SST score obtained from this study was 9.5 points (range being 7-12).

A study conducted by Godfrey et al to measure the reliability, validity and responsiveness of the simple shoulder test showed acceptable psychometric performance. The SST has proven to be useful in the assessment of pretreatment shoulder function along with functional gains and losses over time.

The SST had a good correlation with the SF-12 for physical function. Only 10% of the patients showed either the lowest or the highest scores. The SST was acceptable for most part of the psychometric properties and demonstrated a good test-retest reliability. In comparison to four other shoulder scales such as ASES, SPADI, SSRS, SSI, the construct validity of SST correlated moderately well.

Beaton and Richards, Kirkley et al and Godfrey et al suggested that SST has reliable validity for self-assessment of severity of shoulder function (42). The simplicity of the questionnaire, brief nature and simple scoring property of SST makes it a reliable, valid and responsive criterion to patients' condition. In spite of many advantages, there were few limitations noted. SST was found to be more useful for assessment in older patients as compared to the younger. Subtle changes in function were not recorded due to the yes/ no answer format.

Though reliable and valid, SST did not offer uniform psychometric properties after age group and injury type were stratified. SST was found to be moderately useful in young patients with shoulder instability and is said to assess only the physical functions of the shoulder. Hence, SST is suggested to be used in addition to other measures of assessment like pain and psychological well-being.

MANAGEMENT

Non-surgical management includes exercises, passive stretching, and attempted concealment of the deformity by padding the opposite shoulder. However there was no significant result in improvement by these means.

Most common indications for surgery is mainly cosmetic, and loss of abduction is also an important functional loss. The ideal time for surgery is between the ages of 3 and 8 years, although it can certainly be done after this age but with less satisfying results.

Following factors must be considered in selecting patients for operation

- whether the deformity is on one side or both side
- functional impairment
- cosmetic grade
- age of the patient
- other associated anomalies which may overshadow the shoulder deformity or may not show significant results.

Various methods of surgical treatment are described in the literature. Those are

1. Putti
2. Delchef (1922)
3. Schrock (1926)
4. McFarland (1950)
5. Green (1957)
6. Allan (1964)
7. Woodward (1961)

Initially Putti's method of transplantation of scapula to lower level was by division of muscle attachments and excision of protruding upper part of the scapula. Based on the above technique, various modifications for bringing down the scapula were described by Schrock, Green, Mears and Woodwards (16).

Mcfarland technique was Subtotal scapulectomy - excision of most of scapula, leaving behind only the glenoid and coracoid as he believed that would improve the appearance of the patient. Disadvantages of this procedure were considerable amount of bleeding, incision cannot be planned to give a satisfactory scar and function is often impaired as a result of this surgery.

Putti (1908), Schrock(1926) and Allan (1964) methods of scapular transplantation on the whole have been disappointing. Most often the deformity re-occurs either by return of the scapula back to its previous position or by regeneration of the bone after subperiosteal resection.

Several surgical techniques have been developed and evaluated for this purpose, which can be divided into categories like partial scapulectomy or scapulotomy (25) (43). Scapula translation by replacing the insertions of the shoulder girdle muscles on the scapula or by replacing the shoulder girdle muscles from their origins on the vertebrae. These procedures are performed as a single procedure or in combination with an osteotomy of the clavicle to prevent brachial plexus injury. One of the surgical procedures belonging to the third category was introduced by Woodward in 1961(18). Over the years, several modifications of this technique have been reported. Various procedures including the Putti and Green, Mears and Modified Woodward's procedures have been employed to correct the deformities. Various surgical procedures have been described for elevated scapulae, all offering a reasonable and comparable outcome. In the original Green's procedure, the muscle resection is done distally rather proximally. In this procedure, extensive dissection is required & is technically demanding (44) (45). Modified Green's procedure uses a radiographic geometric method to quantify the lowering & de-rotation of the scapula, but the lowering did not change appreciably with time with the malrotation recurring in 2 years.

GREEN'S PROCEDURE

The Green's procedure is an extensive procedure which involves major resection and also requires a lot of technical expertise (46). There have been a lot of modifications to this procedure by Leibovic and by Belleman to name a few. All the muscles are released extraperiosteally from their insertions. The trapezius is reflected off its insertion medially to expose the Lattismus dorsi, Serratus anterior, Levator scapulae, Supraspinatus and Rhomboids. Following this the supraspinatus fossa is resected carefully to avoid the neurovascular bundle and the omovertebral bone if present is resected. Once the scapula achieves its new position the muscles are reattached and muscle lengthening is performed to maintain the new position. Once the final attachments are made glenohumeral articulation must remain stable and not drift into varus. A velpeau bandage is applied for two weeks and then the child is encouraged to perform pendulum exrecises for several weeks before starting active range of movement exercises.

MODIFICATIONS OF THE GREEN'S TECHNIQUE

LEIBOVIC

He modified the Green's technique by anchoring the scapula in a pocket made in the Lattissmus dorsi muscle (32).

BELLEMANS AND LAMOUREUX

They avoided Serratus Anterior release to enable immediate postoperative mobilization.

ANDRAULT

He advocated osteotomy of the clavicle to prevent brachial plexus palsy.

The main disadvantage of this procedure was recurrence after two years, postoperative winging of the scapula have been reported in few cases and more chances of injury to brachial plexus in older children are also reported. The recommended upper limit for surgery is 8 years of age (44).

Though modified Green's methods have shown good cosmetic and functional results and encourages more surgeons to try this method, it has its own pitfalls. There is an increased chance of keloid formation and postoperative scarring. As mentioned earlier, the Green's procedure has a steep learning curve and involves extensive dissection and requires sound technical skill.

In contrast the Woodward's method has lesser incidence of hypertrophic scars and has produced much better outcomes and has been adopted as the gold standard. This has been quoted in few studies and has produced excellent results. Woodward's procedure is the gold standard with more than 80% having successful cosmetic and functional outcomes.

MEAR'S TECHNIQUE

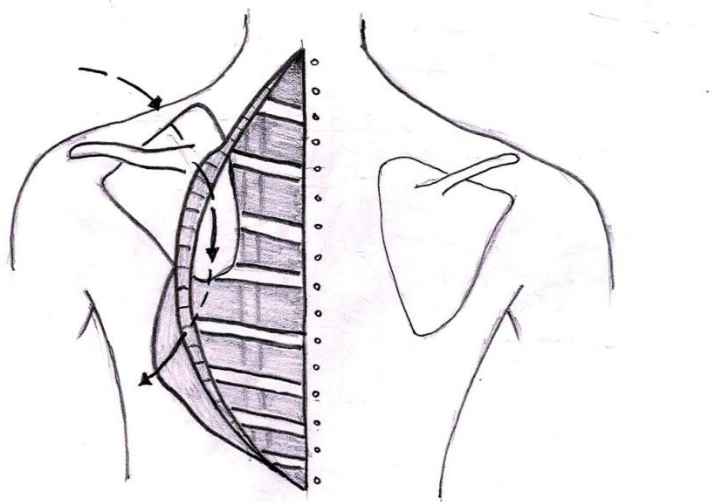
Dana Mears in 2001 described a new surgical technique involving scapular osteotomy, partial excision of the scapula and releasing the long head of triceps. It involves an oblique osteotomy of the supraspinatus fossa to achieve adequate resection of the scapula to prevent impingement (1). Release of the long head of triceps and part of the origin of Teres minor to achieve an increase in abduction range and Infero-medial resection of scapula to enable 160° of shoulder abduction. Clavicle can be excised depending on involvement of brachial plexus. The results from few studies show gradual improvement in range of motion.

Recent reports have shown problems of keloid formation post-surgery, delay in post-operative mobilisation till wound healing occurs and scapular level lowering was less when compared to the modified Woodward's procedure. However, the technique is relatively new and there is very limited literature regarding its application.

WOODWARD'S SURGERY

The Woodward's surgery was introduced by Woodward in 1961 and hence the name (1) (47). The procedure consists of detaching the origins of the rhomboids and trapezius from the spinous process , resection of omo-vertebral bone and fibrous band if present and moving the scapula down(48) (49). Post-operatively, Velpeau sling is used to immobilise the limb and range of motion exercise is begun at 3weeks post-operative.

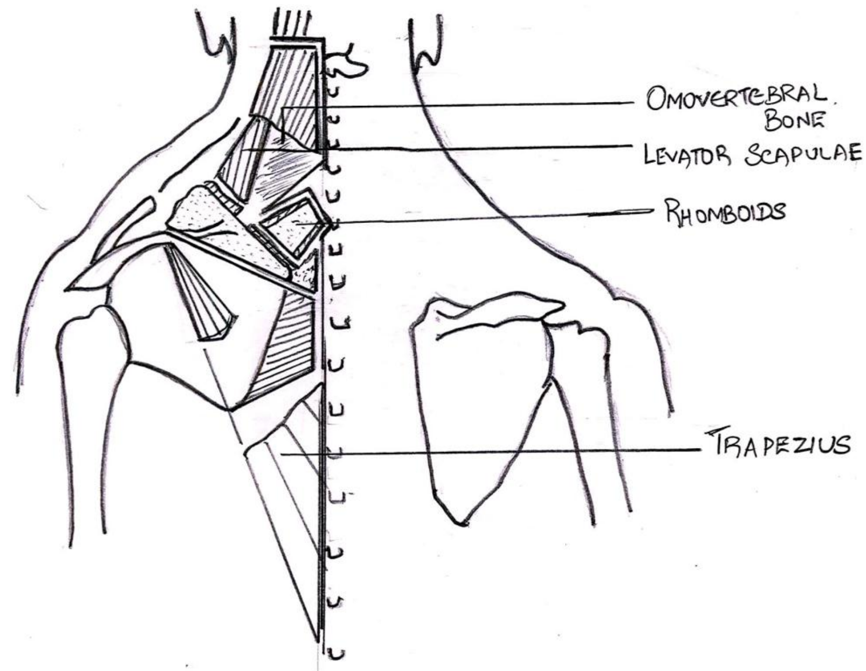
The modification of the Woodward's surgery (6) (14) included addition of excision of the superomedial border of the scapula and resection of the supraspinous portion of the scapula. About 1cm of the medial border of the scapula is excised and the bone resection made superiorly is medial to the scapular notch.



After a midline incision, the Vertebral attachments of the aponeurosis of the trapezoid, rhomboid & levator Scapulae muscles are detached to be able to rotate & relocate the hypoplastic left Scapula more Caudally on the thorax.

→ JSES 2012

Figure 6. Surgical technique by Woodward et al



BEFORE TRAPEZIUS SLIDE

→ STIPPLED AREAS SHOW EXICISED BONE.

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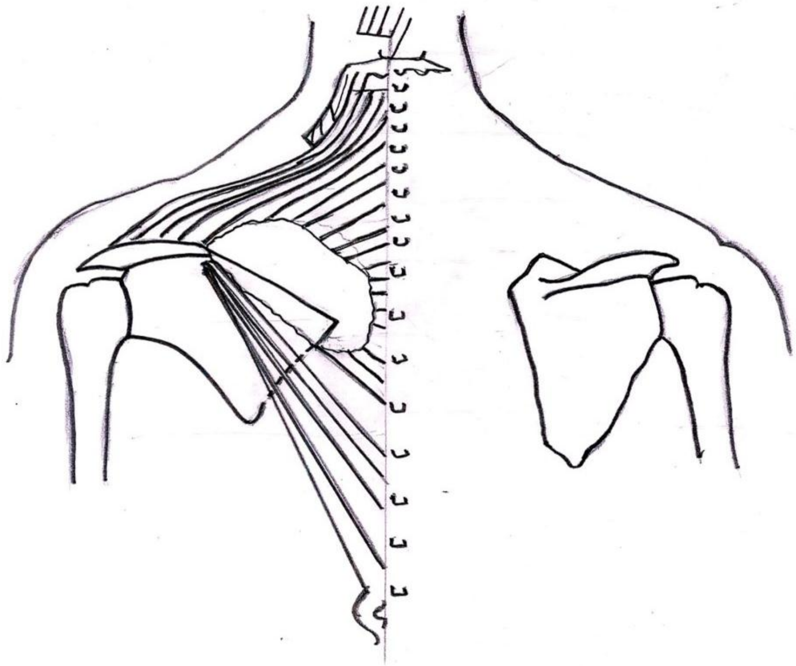
Figure 7. Woodward type scapular transposition



Adopted from the Article:

CONGENITAL ELEVATION OF THE SCAPULA

M.E. CAVENDISH, LIVERPOOL, ENGLAND.

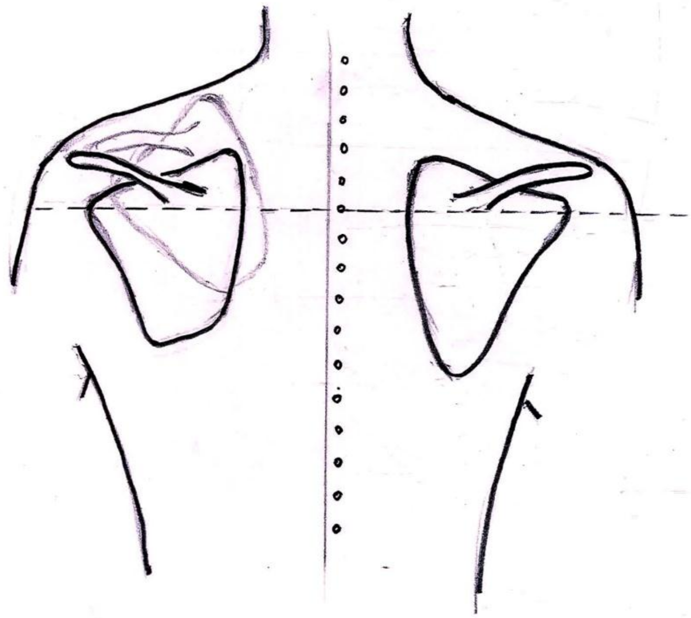
JBJS – VOL 54B, NO. 3 AUGUST 1972

Figure 8. Displacement of scapula after trapezius slide

MODIFIED WOODWARDS SURGERY

Under General Anaesthesia ,the patient is placed in prone position. The arm with scapular gridle are left free for manipulation during the operation. The whole of arm, the shoulder and region from nape of neck from base of occiput superiorly to the lumbar area inferiorly is prepared and draped. A 15cm midline incision is made over the spinous processes extending from the C_4 distally to the D_{10} spinous process.

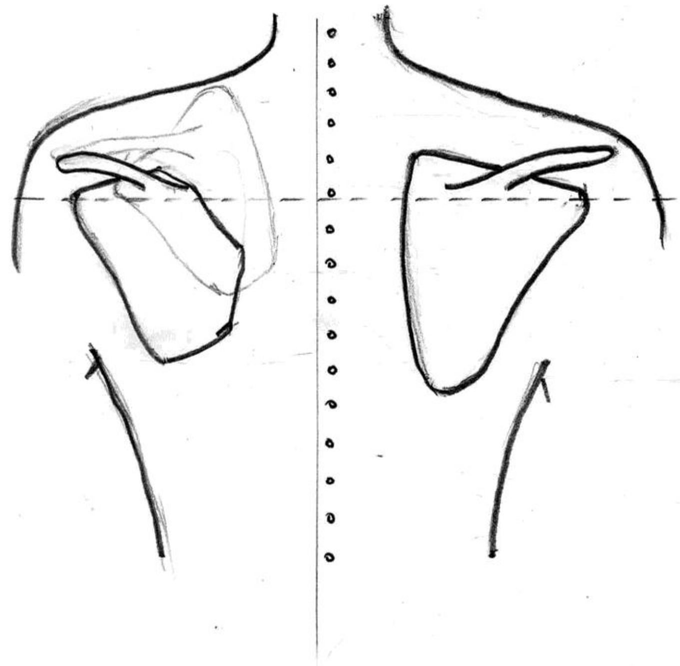
The skin and subcutaneous tissue are undermined laterally to the medial border of the scapula on the involved side and lateral border of the function of the nerves to the arm. Origin of trapezius from C_2 to D_{10} spinous processes are cut and plane between trapezius and serratus anterior is developed. Insertion of Rhomboides is cut close to the scapula. Superomedial border of scapula along with the Omovertebral bone attached to it is removed. Detached insertion of trapezius are sutured back at a more inferior level, pulling the scapula down. The Scapula is reduced, with the trapezius and rhomboids muscle reattached at a more caudal position to the ligaments between the spinous process in the midline. Lattismus dorsi muscle can be lifted to allow the inferior wing of the scapula to be positioned beneath it. The latissimus dorsi muscle is sutured to the inferior tip of the scapular wing. During the reduction, there is a chance that the nerves of the brachial plexus may become entrapped between the chest wall and clavicle. Children who are of higher age and grade -3 Rigualt , clavicular osteotomy is carried out to prevent brachial plexus injury.



The dotted line represents the reference point for the position of the scapular spine of the hypoplastic scapula at the same level as the normal contralateral scapula.

→ Adopted from: JOURNAL OF SHOULDER & ELBOW SURGERY
F.E. WALSTRA, 2012 (1-8)

Figure 9.Surgical technique by Woodward et al.



- Exicision of Supero-medial portion of Scapula along with Omovertebral bone is removed.

The Scapula is reduced, with the trapezius & rhomboids muscle re-attached to more caudal position to the Ligaments between the spinous process at the midline.

Figure 10.Modified Woodward's surgery.

Clavicular Osteotomy:

Incision of about 2cm is made over the mid clavicular region. Plane is created beneath the platysma muscle, the periosteum is incised longitudinally and sub-periosteal elevation exposure of the clavicle is done. Osteotomy of the clavicle is carried out and rongeur is used to make chips of cortical bone over the mid-area of clavicle for a length of approximately 1cm. The clavicular osteotomy is required in case of older children or with associated deformity. The wound is closed in layers over a suction drain. Sometimes VAC dressing is applied based on surgeon's preference.

COMPLICATIONS OF SURGERY

Complications following surgery include nerve palsy, brachial plexus injury, incomplete correction, recurrence, wound infection and operative scar appearance. The brachial plexus is at risk of compression intraoperatively as the scapula is displaced inferiorly. Several authors have reported a 6 to 11% brachial plexus palsy after surgery for Sprengel's deformity and tended to occur in severe deformity and older children; however, many palsies were transient. Clavicular osteotomy should be considered for brachial plexus injury prevention in these groups. Intraoperative somatosensory-evoked potential monitoring may help to prevent such injury. In literature the prevalence of Hypertrophic scar was 26–64%, Regrowth of the superior pole of the scapula was 30% andScapular winging 4–17%.(3)

The modified Green technique has been reported to cause keloid formations because of the site of incision over the shoulder, recurrence of the scapula elevation after 2years and postoperative winging of the scapula in some cases.

Presently, the preferred method is Modified Woodward’s procedure since it does not require a postoperative spica cast for suture anchor, entails a lower risk of bleeding & brachial plexus injury and has shown good results in the past. This procedure has got advantages over the Putti-Schrock-Green methods of scapular transplantation and is less likely to cause a spreading scar. Since the incision is in the midline, post-operative scapular fixation by scarring is less likely since the dissection around the scapula is less likely and recurrence of the deformity is less likely.

METHODOLOGY

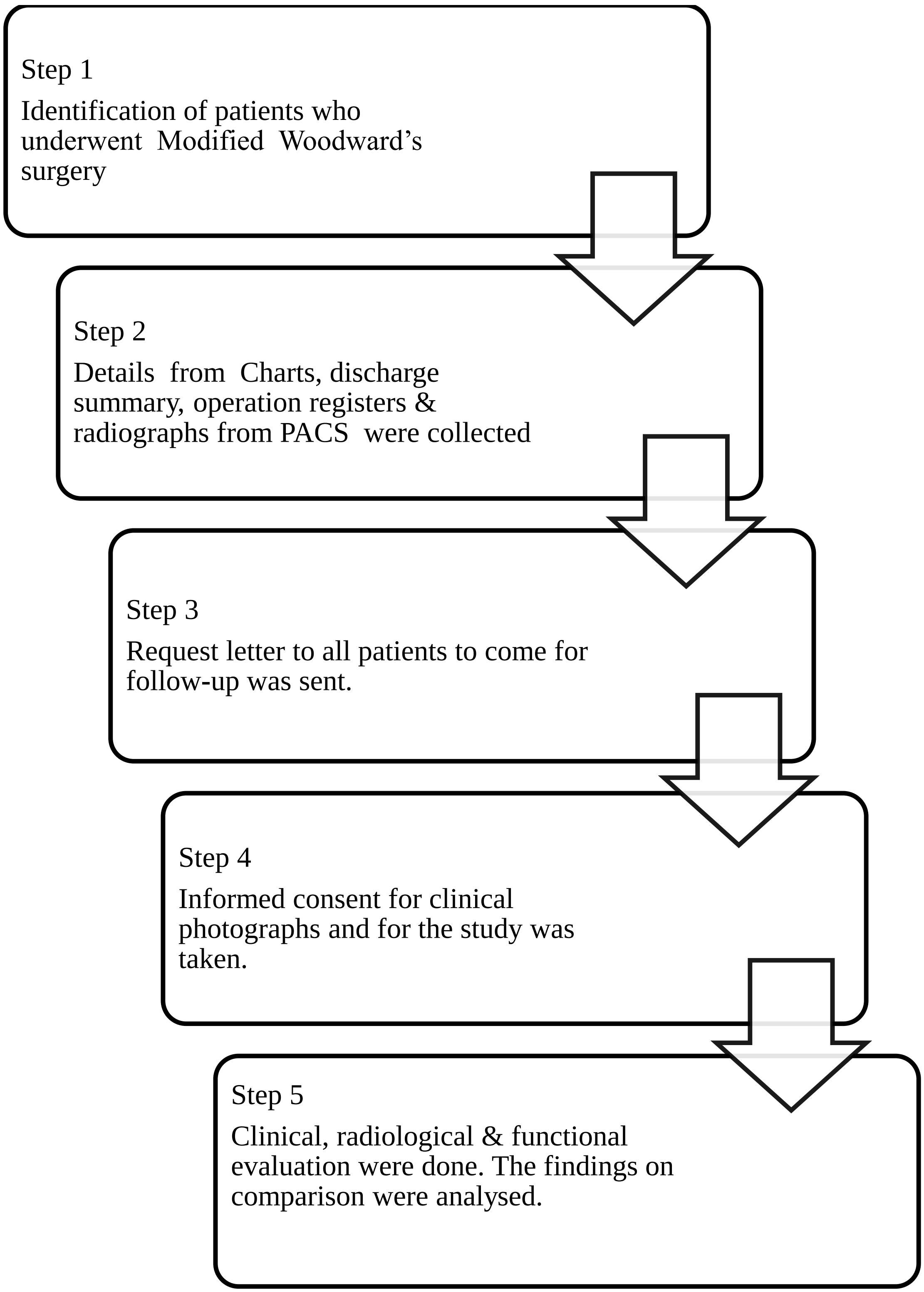
In this retrospective observational study, twenty two patients with Sprengel's deformity who underwent Modified Woodward's procedure with a minimum of one year follow-up were included and examined. The patients who underwent surgery during the period of January 2006 – August 2014 under the Paediatric Orthopaedics unit in CMCH , Vellore were included in this study.

Sprengel's deformity children who underwent Modified Woodward's procedure between 2006-2014.

INCLUSION CRITERIA

- Age: Children operated below 18 yrs of age.
- Operation date: between January 2006- August 2014.
- Center where operated: Children who underwent Modified Woodward's procedure in the Paediatric Orthopaedics unit in CMCH , Vellore.

STUDY ALGORITHM



Preoperative findings were noted including patient's detailed history, examination findings, the family history of a similar problem, other skeletal anomalies like Scoliosis, Omovertebral bone, Klippel-feil syndrome, Congenital Vertebral anomalies like anterior chest wall deficiency, rib anomalies and any problems during gestation were recorded. The operative indications were divided into one of these 3 categories

1. Severe restriction of range of abduction at the shoulder,
2. Interfering with activities of daily living like difficulty in dressing oneself or
3. A cosmetically displeasing deformity, or both.

Associated musculoskeletal abnormalities like Omovertebral bone, Klippel-Feil syndrome, scoliosis, spina bifida & rib anomalies were also noted.

Cavendish grading was used to evaluate cosmetic appearance. Range of motion, Paediatric outcomes data collection instrument (PODCI) and simple shoulder test (SST) were obtained to evaluate shoulder function. Scapula placement and degenerative disease were assessed by radiographic examination and by the Rigault's classification.

Examination of the shoulders, both upper limbs, spines, and lower limbs were done with giving special attention for scapular bony prominences, scapular winging, scoliosis, chest asymmetry, or any associated morphological abnormalities. All the patients were examined for the grade of the deformity. Movements of the shoulders were examined as well as of elbow, wrist, and hand; Range of motion especially of the scapulo-thoracic motion was noted, to determine whether the scapula was anchored to the spine or not.

Standard radiographs of antero-posterior view of both shoulders or Chest X-Ray were done, few needed radiographs of the cervical, thoracic, and lumbar spines in patients who have associated anomalies. Measurments of Superior displacement ratio was done by drawing two reference lines. Line 1 was drawn from the center of the glenoid cavity of the affected shoulder perpendicular to the vertebral axis line, and line 2 was drawn from the normal shoulder. The superior displacement ratio was formed by the distance between these two lines as a fraction of the scapular height on the normal side. This ratio is not measured in bilateral cases, as there is no normal side to use as a reference.

To evaluate the height-to-width ratio, the height of the scapula was measured from the superior angle to the inferior angle parallel to the glenoid, and the width was measured from the cavity of the glenoid to the most medial portion of the vertebral border perpendicular to the glenoid on the radiograph. To measure scapular descent after surgery, the inferomedial angle of the scapula was used as the reference point for evaluation. Associated spinal anomalies, scoliosis and hemivertebrae were also documented post-surgery.

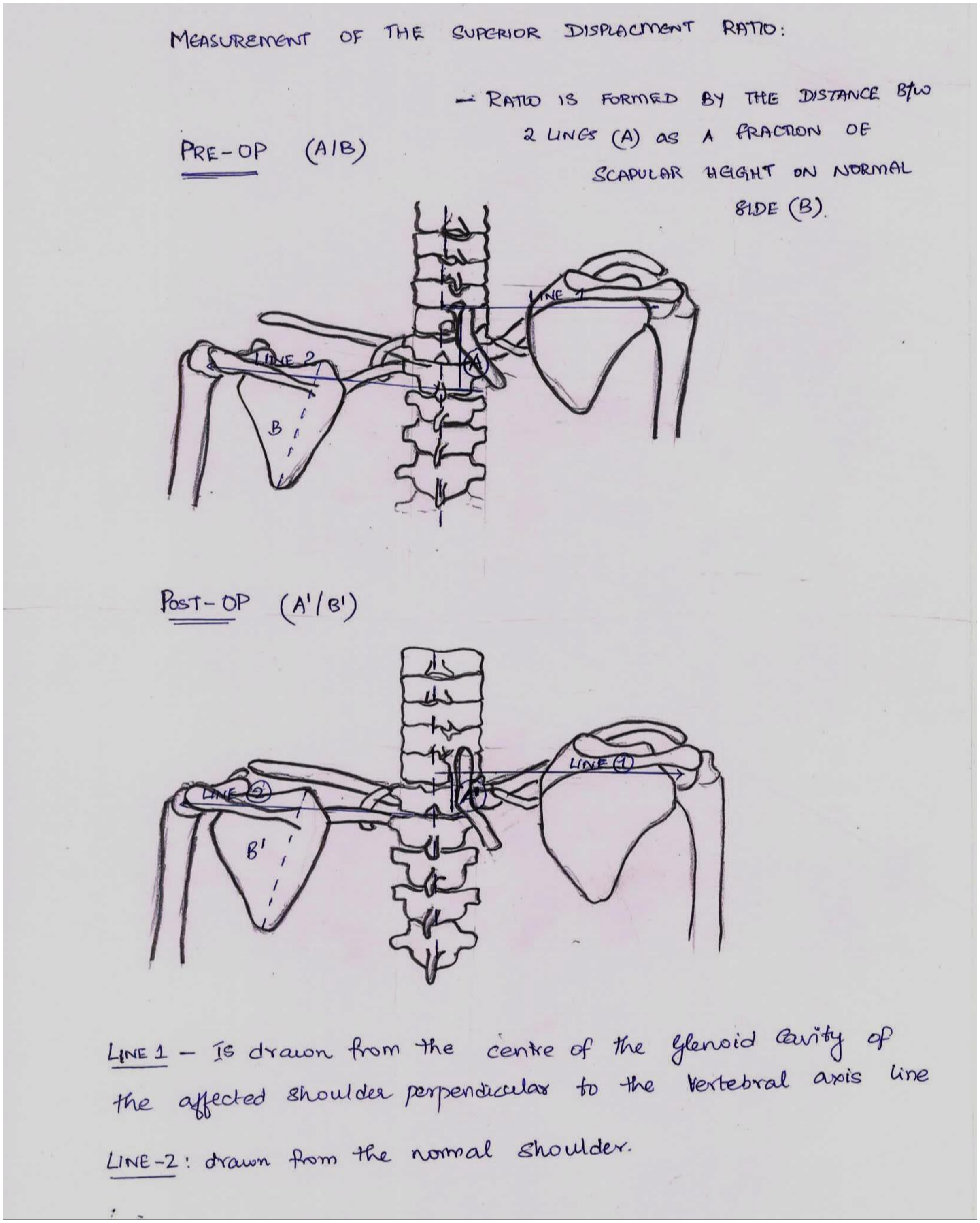


Figure 11.Measurement of the superior displacement ratio.

The Cavendish score and Rigault's radiological score was used to assess the severity of the deformity & the position of the scapula relative to the cervical spine respectively and functional score were also assessed with Paediatric outcomes data collection instrument (PODCI) and simple shoulder test (SST).

On follow up, the data was evaluated for deformity using the Cavendish grade as a measure of cosmetic improvement and rangeof abduction at the shoulder as a measure of function.

Table 2. Cavendish Score

Cavendish Score	**Sprengel's deformity**
Grade 1 - Very mild.	Shoulder joints are level. Deformity is invisible when patient is dressed.
Grade 2 - Mild.	Lump in the web of the neck. Deformity is visible when dressed.
Grade 3 - Moderate.	Shoulder elevation 2-5 cm. Deformity is easily visible.
Grade 4 - Severe	Superior angle of the scapula is near the occiput with/without neck webbing or brevicollis

Radiological evaluation was performed using the Rigault's classification. This classification is based on the location of the superior-medial angle of the hypoplastic scapula.

Table 3.Rigualt's classification

Rigault's Classification	
Grade 1	Under Thoracic-1
Grade 2	between Th1, and C5
Grade 3	above C5.

1. The PODCI scoring system has 5scales & one global scale. It has a separate eight-item questionnaire for the upper extremity & physical function which includes the standardized score. A score of "0" represents a poor outcome while "100" is best possible outcome. A minimum of 4 items must have valid answers to score this scale through raw scores & mean of items. Adolescent (parent-report) Outcomes Questionnaire is intended for youth in the age group 11 to 18 who are capable of completing the form independently. This questionnaire is similar to the Adolescent (parent-report) Outcomes Questionnaire, but does not offer a response option indicating that the respondent is "too young for this activity."

2. The SST consists of 12 questions with dichotomous response options. For each question, the patient indicates whether he or she is able to do the activity or not. The scores are summarized into a total score, which ranges from 0 (worst) to 12 (best) for shoulder functioning. Missing data was treated as follows: 1 or 2 missing values will be substituted with the average value of the other items. If more than 2 items are missing, the response to this questionnaire was considered invalid and no total score was calculated. The maximum score of the SST is 12 points, higher values indicating better shoulder function. Furthermore, at the long-term follow-up examination, patient or parents were asked to rate the overall satisfaction in terms of bad, fair, or good.

PROFORMA FOR DATA COLLECTION

A proforma for evaluating the different aspects of the patient was filled up and entered into the Microsoft Excel spread sheet which was used for analysis. Proforma enclosed in annexures.

STATISTICAL ANALYSIS

Statistical analysis was done using STATA v.13 software. Paired t-test was used for the paired analysis of preoperative and final values of Cavendish grade, range of abduction, ratio of superior displacement and height width ratio. The Rigault grade and Forward flexion are not normally distributed population; we therefore used the Wilcoxon signed rank test. Impact of age group and associated malformations on the end result was evaluated by the Student t-test and Mann-Whitney test for normal and skewed data respectively. Mean and SD reported for normally distributed data and Median (IQR) for non-normally distributed data.

RESULTS

Out of twenty two children who underwent Modified Woodward's surgery, only fourteen patients were available for follow- up. No patients had prior shoulder surgery.

Out of twenty two patients who underwent modified Woodward's surgery for Sprengel's deformity. Fourteen patients responded to come for follow-up, two patients were not willing to participate in the study, three patients responded stating that they will be coming for follow-up during exam holidays, three patients were lost to follow-up. Out of two patients who were not willing for the study, one had brachial plexus neuropraxia which was operated at age of five. She was taken back to OT and underwent suture release on the same day. The other patient was doing well in her activities of daily living without any problem but wasn't willing to participate in the study.

Out of fourteen patients, three were boys, eleven were girls. They were operated at a mean age of 5.7 years (range 3-12). Four patients had been operated on right side, 9 patients on left & one underwent bilateral correction. Six patients had associated anomalies, with one having Klippel-Feil syndrome, four had scoliosis, and one had scoliosis, with congenital vertebral anomalies and anterior chest wall deficiency.

In the patient with bilateral deformities, the Right side was operated first followed by the left side six months later. Ten patients out of fourteen had an Omo-verterbal bar, with the bilateral patient having it on both sides. The superomedial angle of scapula excision was performed in all shoulders.

Two patients had unsatisfactory cosmetic results due to co-existent conditions like Klippel-Feil syndrome & congenital vertebral anomalies with anterior chest wall deficiency.

One patient had stitch granuloma on his back, 5 months post-surgery, which was removed. Following this, he did not have any complication or scar hypertrophy.

No patients had surgical scar complications like wound necrosis or keloid formation. Hypertrophic scar formation was seen in two patients.

Cavendish Grade-4 deformity was present in 6 patients & 7 patients with Cavendish Grade-3. One patient had bilateral grade-3 involvement. According to Rigualt's classification - 7 shoulders had grade-3 and 6 shoulders had grade 2. The patient with bilateral involvement had grade-2 both sides.

Mean follow up was 54 months with the mean abduction of the shoulder improved from 107.1^0 preoperatively (90-130) to 143.57 post-operatively ($100\text{-}170^0$). The mean forward flexion* improved from 120.0 preoperatively (100.0,130.0) to 160.0 post-operatively (150.0,160.0). The abduction range of movements had an improvement of 36.5^0 with significant increase ($P<0.001$). The forward flexion also had a statistically significant improvement ($P=0.0008$).

There was cosmetic improvement by Two Cavendish Grades in 14 shoulders. The preoperative Cavendish grade mean was 3.43 to post-op 1.42 with a significant P-value <0.001.

Radiological improvement of scapular lowering was assessed by Rigualt's grade* which showed preoperatively mean 2.5 (2.0, 3.0) to post-op 1.0 (1.0, 1.0) grade with significant p-value of 0.0006.

Other radiological assessment such as Superior displacement ratio had significant improvement (p=0.0001) (pre-op-0.40 compared to post-op of 0.28). Height/width ratio showed statistically significant improvement(p-value of 0.008). No signs of degenerative changes were found in shoulder joints at follow-up radiologically. The mean scapular lowering was 2.01 cm.

Evaluation of questionnaires with PODCI score & SST was done at follow-up, showed mean PODCI Score of 83.21 (Range – 58-100) & mean SST score of 9.6 points. (Range 8-12). Most patients had encountered functional disabilities like while lifting heavy object above shoulder height with left hand for respect to their age. Parents & patient's satisfaction concerning the results of the Modified Woodward's procedure was rated good at follow-up in all patients.

Comparison of outcome was done in two subsets group of patients

1) patients with age <7 years and more than 7 years at time of surgery
2) Patients with associated anomalies versus patients with no associated anomalies.

Each group was compared with Cavendish grade, Rigualt grade, Abduction, forward flexion – range of movements, height width ratio & PODCI score.

There was significant improvement in forward flexion in patients of age <7 yrs with p-value 0.03.

The patient with no-associated anomalies [1.75(.46)] had significant improvement in Cavendish score p-value 0.046 when compared to those with associated anomalies [2.3(.52)]& Rigualt's grade with p-value of 0.008 respectively. All other factors between the groups did not show significant values.

Note: Values are reported as mean (SD) for normally distributed variables * median (IQR) for skewed variables.

Table 4.Comparison of variables pre-test and post-test.

Variables	**Pre test Mean (SD)**	**Post test Mean (SD)**	**p-value**
Cavendish grade	3.43 (0.50)	1.428 (0.51)	<0.001
Rigault grade*	2.5 (2.0, 3.0)	1.0(1.0, 1.0)	0.0006
Abduction (Degree)	107.1 (17.72)	143.57 (16.45)	<0.001
Forward Flexion*	120.0 (100.0, 130.0)	160.0 (150.0, 160.0)	0.0008
Superior displacement	0.40 (0.09)	0.28 (0.07)	0.0001

ratio			
Height/width Ratio	0.90 (0.10)	0.95 (0.07)	0.008

Table 5. Comparison of variables based on age

Variables	<7 years Mean (SD)	>=7 years Mean (SD)	p-value
Cavendish grade	2 (0.5)	2 (0.7)	0.999
Rigault grade	1.0 (1.0, 2.0)	1.0 (1.0, 1.0)	0.61
Abduction (Degree)	-38.9 (9.3)	-32 (8.4)	0.194
Forward Flexion	-40.0(-60.0-30.0)	-30.0(-30.0-30.0)	0.03
Superior displacement ratio	.116 (.09)	.116 (.06)	0.995
Height/width Ratio	-.043 (.07)	-.052 (.03)	0.792

Table 6 Comparison of data in patients with associated anomalies and patients without associated anomalies.

Variables	Associated anomalies Mean (SD)	Non-Associated anomalies Mean (SD)	p-value
Cavendish grade	2.3 (.52)	1.75 (.46)	0.046
Rigault grade	2.0 (1.0, 2.0)	1.0 (1.0, 1.0)	0.008
Abduction (Degree)	-31.6 (11.7)	-40 (5.3)	0.097
Forward Flexion	-35.0 (-40.0, -30.0)	-30.0 (-55.0, -30.0)	0.94
Superior displacement ratio	.091 (.09)	.137 (0.05)	0.287
Height/width Ratio	-0.045 (0.02)	-0.048 (0.07)	0.937

Note: Values are reported as mean (SD) for normally distributed variables * median (IQR) for skewed variables

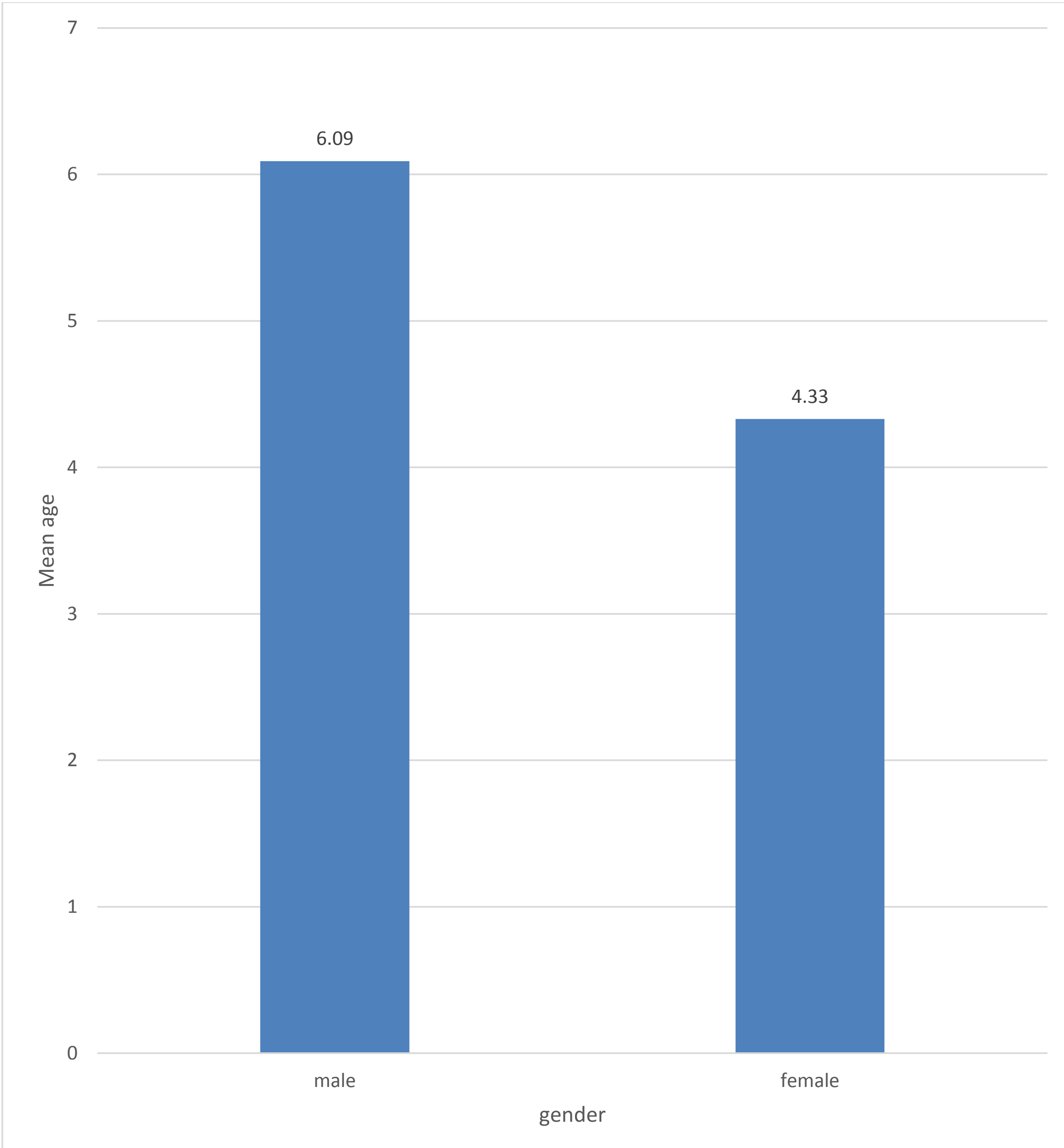


Figure 12. Graph showing Age and sex distribution of the study population

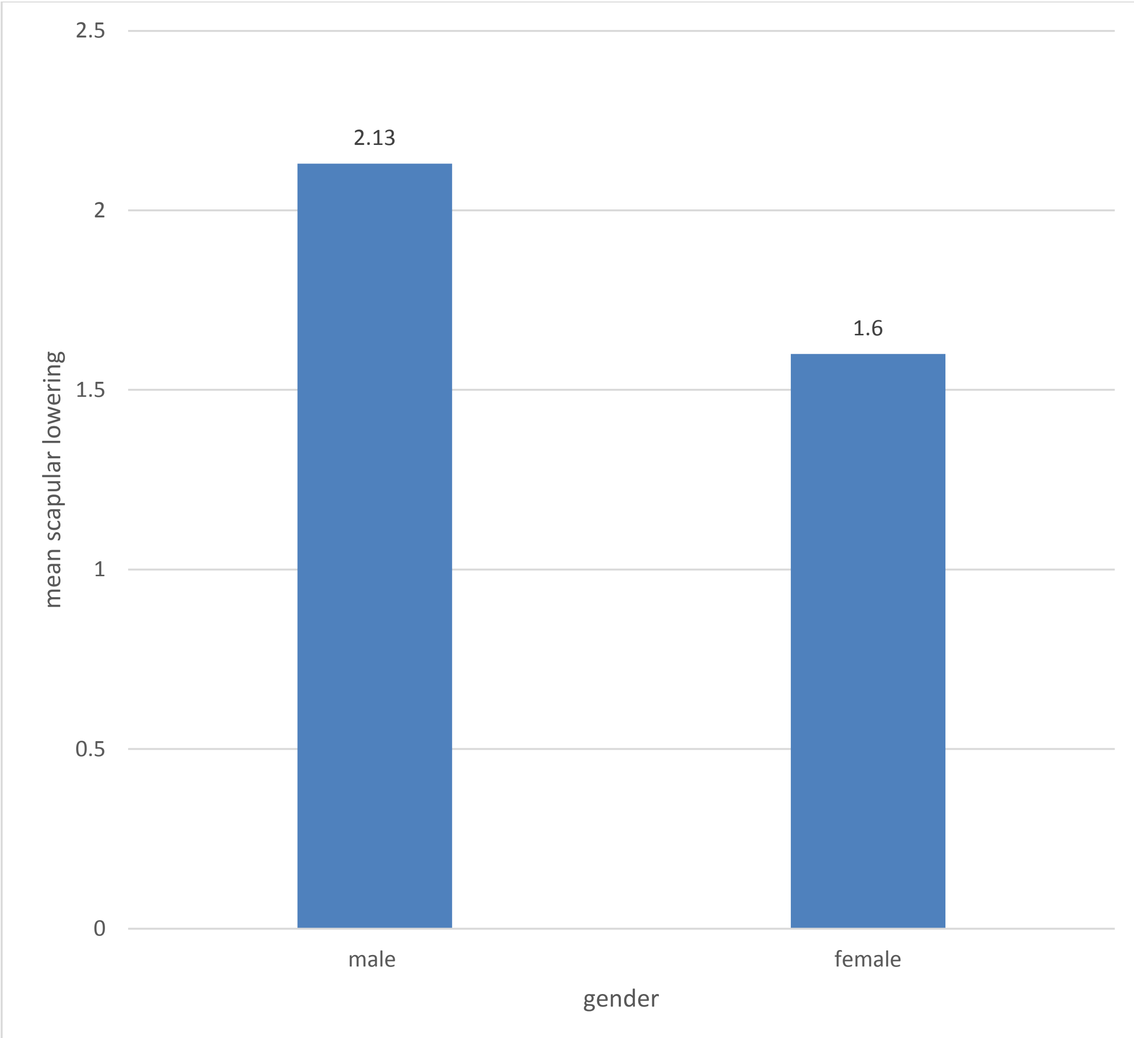


Figure 13. Shows mean Scapular lowering between males and females

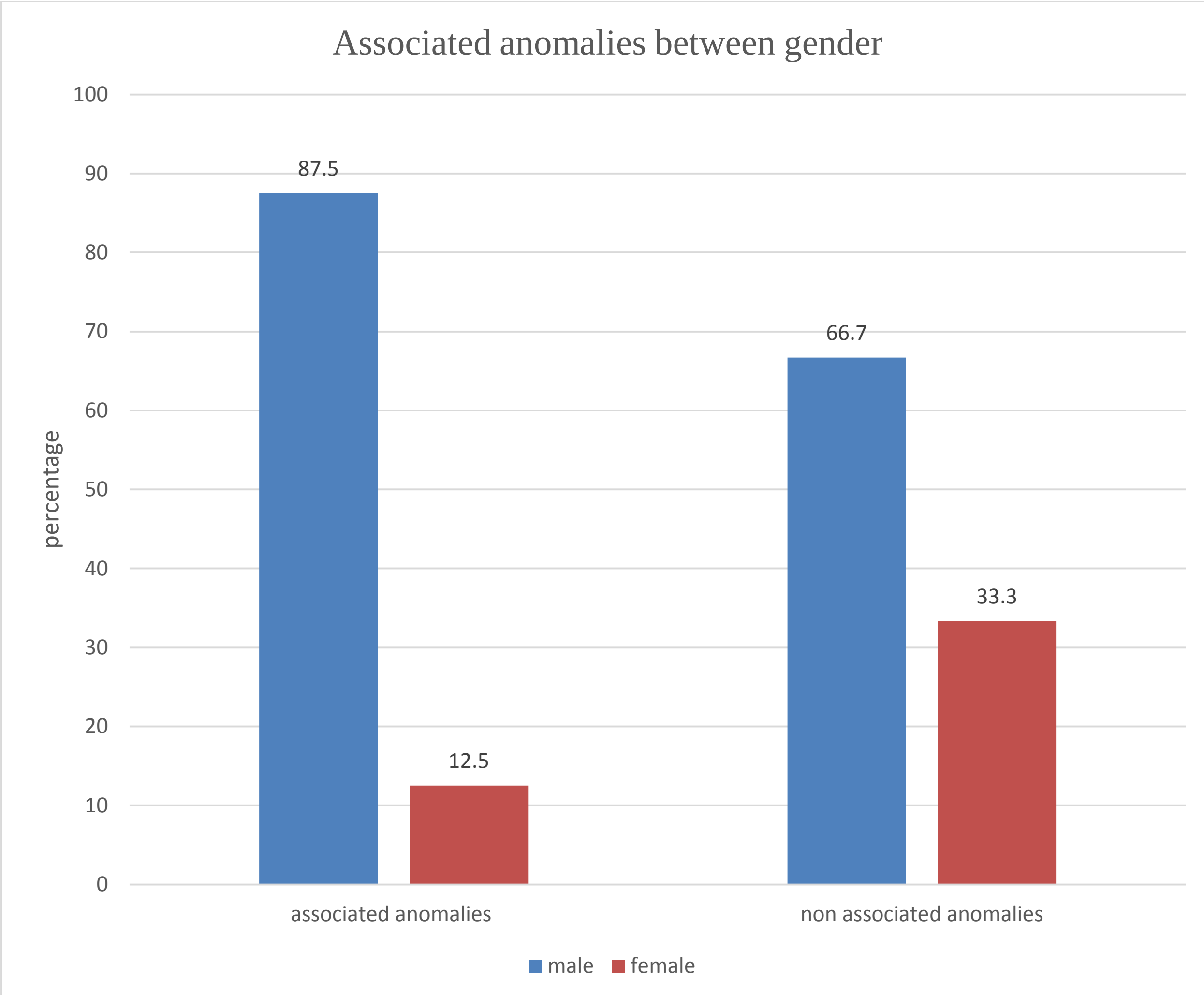


Figure 14.Shows associated anomalies between male and female patient in this study

DISCUSSION

Sprengel's deformity is a congenital elevation of the scapula during embryonic development. In a growing child, the impairment is static since the fibrotic tissue does not grow(18). The deformity can cause severe impact on activities of daily living and shoulder function(18). Limited shoulder abduction and cosmetic appearance are the major concerns. Non-operative management has no positive outcome in treatment of Sprengel's deformity (13) (34) (46) (50). There are a number of surgical procedures described to correct the above deformity. Of the procedures described like Woodward's, Green's, Mears' and their modifications, the modified Woodward's procedure has been found to be the most commonly used. Though many studies are reported, very few have described the functional outcome of surgical correction of Sprengel's deformity.

In this study we used the Cavendish score for assessing cosmetic improvement, Rigault's score, height-width ratio of the scapula and superior displacement ratio for assessing radiological improvement, PODCI (Paediatric outcomes data collection instrument) and simple shoulder test (SST) scores for assessing functional improvement.

The Cavendish and Rigault scores are widely used in literature (3)(33)(18) and form a common platform for comparing outcomes between studies. These were used by us because of their ease of use, and because they assess more exactly the amount of deformity and improvement post operatively. Gupta et al (12) reported on twelve children who underwent the Woodward's procedure at mean age of 5.8 years, mean pre-operative

Cavendish score of grade-3.17 & range of abduction at the shoulder of a mean of 115.83°. At an average of 31.8 months of follow-up, there was statistically significant improvement to a Cavendish grade of 1.25 and abduction range of 153°. Similarly Walstra et al (18) reported on 8 shoulders at an average of 14.7 years which showed cosmetic improvement from pre-operative Cavendish grade 3 to grade 1 or 2 at final follow-up. In our study the cosmetic improvement by an average of 2 Cavendish grades (from 3.43 to 1.42) in the 14 shoulders assessed with a significant P-value <0.001 is higher than reported. One of the main reasons for this is a generous resection of supero-medial corner, an essential component of the modified Woodward's. When comparing with the modified Green's, long term results reported by Gonen et al (51) in 24 children showed improvement of at least one Cavendish grade in 88.9% of shoulders. Mear's procedure outcomes have focused mainly on the improvement in range of motion and a cosmetic comparison is not possible based on the existing literature.

Radiological improvement of scapular lowering was assessed by Rigualt's scoring system again a commonly used outcome measure for this modality. Our study showed improvement of more than 1grade (median of 2.5 to 1) post-operatively with a significant p-value of 0.0006. Other radiological assessment such as Superior displacement ratio had shown significant improvement (p-value of 0.0001) in our population. Walstra et al(18) using Woodward's procedure showed a 17.8% significant improvement of one grade in 8 shoulders. Improvement of Height/width ratio in our study was small but significant with a p-value of 0.008. The mean scapular lowering in our study was 2.01cm which is comparable to the value of 2.2 cm reported by Siu et al (33).

Comparison of outcome was also assessed in patients with associated anomalies vs no associated anomalies. The patient with No-associated anomalies had significant improvement in Cavendish & Rigualt grade with p-value of 0.046 & 0.008 respectively. All other factors between the groups did not show significant values. This supports the established practice of either avoiding surgery or doing surgery with counselling of the patient in terms of cosmetic improvement when associated abnormalities are present. Range of motion is an important factor in shoulder function and has been specifically targeted in surgery that is described by Mears. There was a significant improvement in abduction and forward flexion in our patients. This was more so significantly in forward flexion range (a mean improvement of 40°). This was significantly more in children under the age of seven years. Earlier studies on the results of the Woodward procedure reported a mean functional improvement in abduction of 32-59° and few complications. The improvement in shoulder abduction in our series was in concordance with these results. Our study showed an improvement in abduction range by a mean of 36.5^0($P<0.001$) at mean follow up of 54 months. Other series on Woodward's such as by Walstra et al and Mushin et al (32) also showed improved range of motion in the long term. Mears et al showed a higher range of abduction improvement at a mean of 60° in 8 patients as compared to Woodward's procedure (47) .Mears is a very major procedures involving release of triceps from the glenoid and an osteotomy through the body of the glenoid and as functional improvement is not shown to be superior it is debatable if an extensive procedure is warranted over Woodward's.

When we combined our data with the eligible published data concerning abduction, the combined data of 14 cases confirmed a significant abduction, forward flexion, scapular displacement and height width ratio significant improvement following surgery and at follow-up.

Leibovic et al (32) modified the Green technique by suturing the reduced scapula into a pocket formed in the Latissimus dorsi muscle. In 16 shoulders (14 patients), at a mean follow-up of 6.5 years, mean shoulder abduction was reported to be148° (range, 100° to 180°), compared with 91° preoperatively (range, 60° to 120°). Although the initial results were encouraging, scapular rotation was not maintained at longer-term follow-up and half the cases developed hypertrophic scarring.

The quality of life and functional outcomes following Sprengel's deformity surgery have been measured using scoring systems like Constant Score, DASH(Disability of Arm, Shoulder and Hand Score) and SST (Simple Shoulder Test). The PODCI score has previously been reported to be a valid and reliable instrument for assessing the function in conditions such as cerebral Palsy, amyoplasia congenita and unilateral upper extremity deficiencies(U-UED) in comparison with a group of normal children. To our knowledge PODCI scoring system has never been used before for studying the outcome of Sprengel's deformity. . The SST, on the other hand has been used for Sprengel's deformity and found acceptable with demonstrated good test-test reliability and validity. In comparison to four other shoulder scales such as , American Shoulder and Elbow Surgeons Standardized Shoulder Assessment, Shoulder pain and disability index, SSI, the construct validity of SST correlated moderately well.

Some of the studies evaluating Sprengel's have evaluated function using Constant Score and DASH (Disability of Arm, Shoulder and Hand Score). Constant score is not originally meant for assessing children, not standardised for children and the strength component has significant variability and unreliability with varying ages and sex and not a good outcome tool. To our knowledge, the PODCI score has never been used in literature to assess functional outcomes for Sprengel's deformity. The scoring system is very easy to use and reliable. We chose PODCI and SST scoring systems to assess the functional outcomes. A mean PODCI Score of 83.21 & mean SST score of 9.7 points was found in our patients. The maximum score of SST was fixed as 12 points. Better shoulder function had higher values. The mean SST score obtained from the study conducted by Walstra E F et al(18), for Sprengel's deformity patients corrected with Woodward procedure was 9.5 points (range being 7-12). The findings of the prospective study by Roy et al showed that the clinical importance difference of SST is 3 on the 12 point SST scale. Parents & patient's satisfaction concerning the results of the Modified Woodward's procedure was rated good at follow-up in all patients. The PODCI score of 83.21 was considerably higher than the normative score of 50 indicating that the function in these children was above average and not compromised.

Complications which were reported in earlier series included scar dehiscence, scapular winging, brachial plexus injury and long term complications like keloid formation and unsightly scars. The brachial plexus is at risk of compression intraoperatively as the scapula is displaced inferiorly. Several authors have reported a 6 to 11% brachial plexus palsy after surgery for Sprengel's deformity and tended to occur in severe deformity and older children; however, many palsies were transient. Clavicular osteotomy should be considered for brachial plexus injury prevention in these groups. Intraoperative somatosensory-evoked potential monitoring may help to prevent such injury.

In literature the prevalence of Hypertrophic scar was 26–64%. In this study we had one patient with brachial plexus injury and two with hypertrophic scars. Earlier studies have reported the best time for surgery is between 3-8 years of age as at an older age clavicular osteotomy to prevent brachial plexus injury is required and degree of correction is less. In this study, however, there were 5 patients above 7years who underwent surgery and showed results almost equal to the patients less than 7years in all parameters other than forward flexion.

Compared to Woodward's the modified green technique has been reported to cause keloid formations because of the site of incision over the shoulder, recurrence of the scapula elevation after 2years and postoperative winging of the scapula in some cases. In terms of keloid formation risk and postoperative scarring, better results were reported using the Woodward's method than Green's by Greitemann B et al (1993) (23) in treatment of congenital elevation of the scapula in follow-up of 37 cases of Sprengel's deformity.

In conclusion our study showed significant improvement in cosmesis, radiology range of motion, scapular position and function using modified Woodward's procedure. The results obtained were in concordance with the published literature. It is a safe and predictable method with very few complications.

Figure 15.13years old girl who underwent modified Woodward's surgery bilaterally at age of 5yrs with pre-op abduction of 120° and forward flexion of 130° bilaterally.

Figure 16.She had abduction of 160° and forward flexion of 170° bilaterally post-operatively

Figure 17

Figure 18

Her post-op PODCI scoring was 92 on right side and 80 on left side.

Figure 19

Figure 20.18years old girl who underwent modified Woodward's surgery on left side at age of 2yrs with pre-op abduction of 110° and forward flexion of 130°

Figure 21. She had abduction of 150° and forward flexion of 160° post-operatively

Figure 22.15years old girl who underwent modified Woodward’s surgery on left side at age of 8yrs with pre-op abduction of 130° and forward flexion of 140°

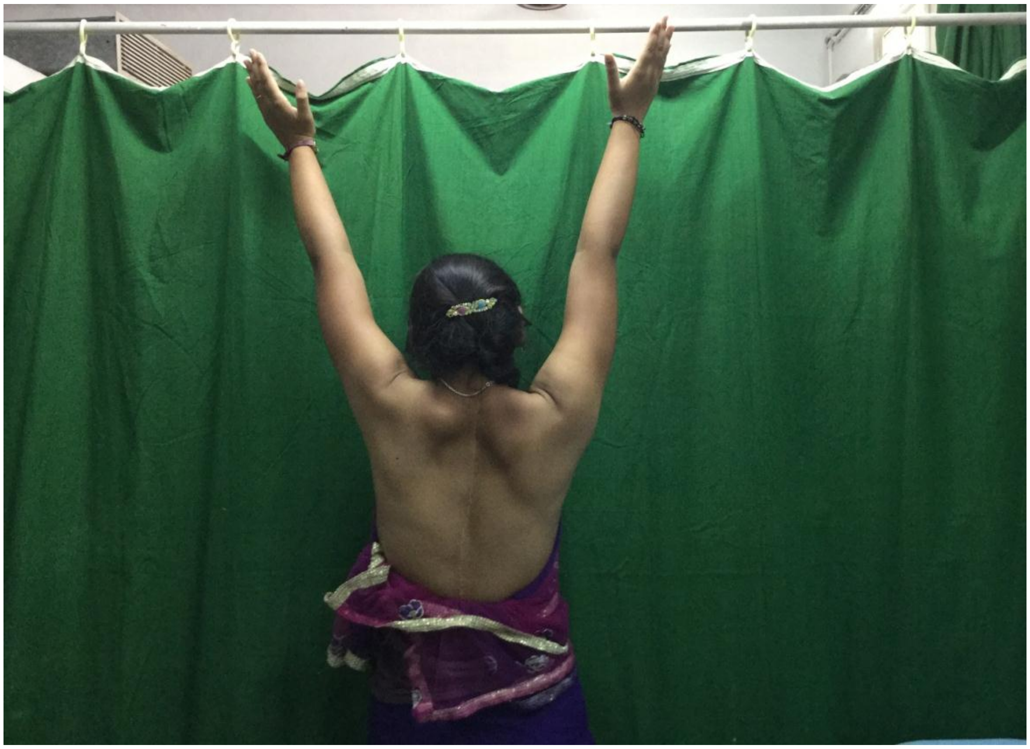


Figure 23.She had abduction of 160° and forward flexion of 170° post-operatively

Figure 24. She had Cavendish score of grade -4 pre-op to Grade- 1 post-operatively. Her PODCI Score was 100.

Figure 25.7 Years old boy who underwent modified Woodward’s procedure with congenital vertebral anomalies and right anterior chest wall deficiency

Figure 26 He had pre-op Abduction of 60° and forward flexion of 80°

Figure 27 Post-op he had Abduction of 100° and forward flexion of 120°

Figure 28. Cavendish grade -4 pre-op to grade -2 postop

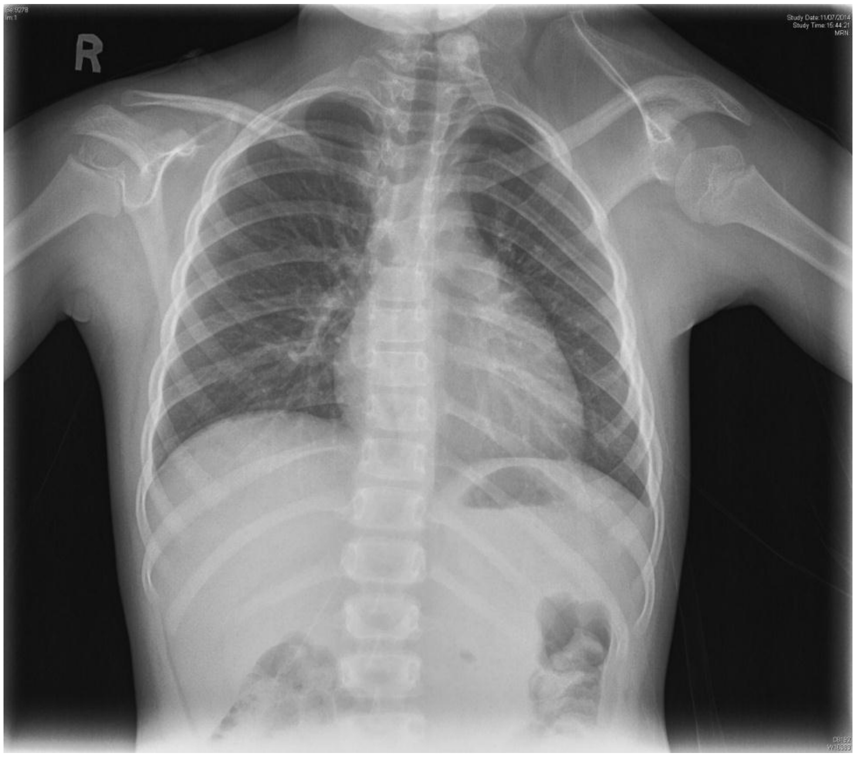


Figure 29.X-ray showing elevated scapula on left side pre-op with Rigualt's grade-2 of a 3years old child

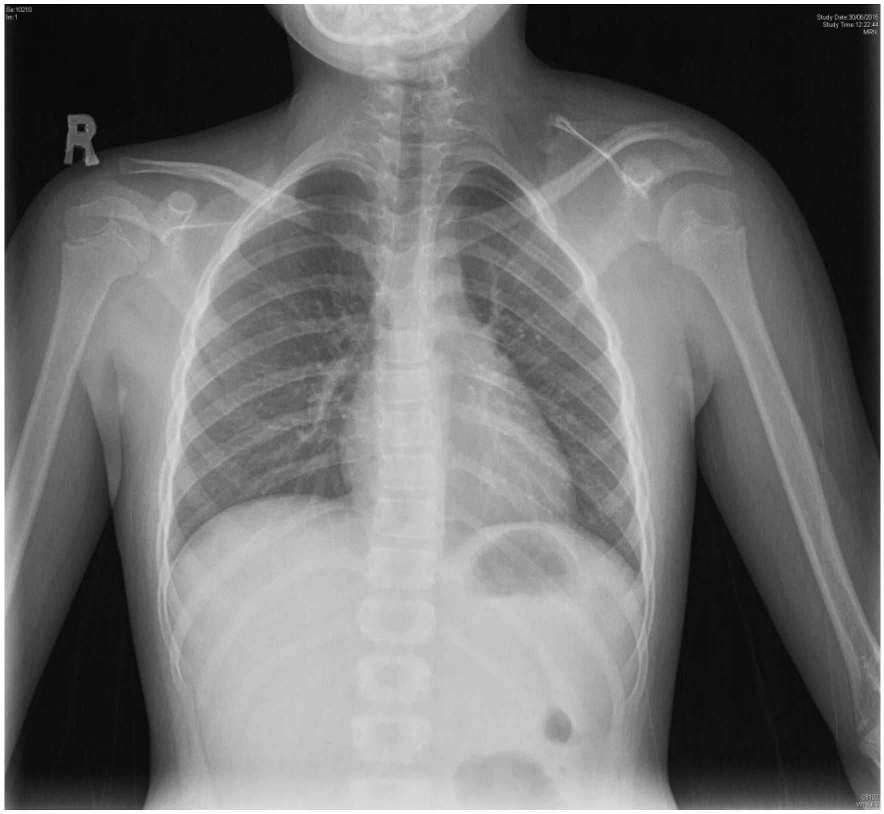


Figure 30. Post-op X-ray showing Rigualt’s grade-1 at follow-up

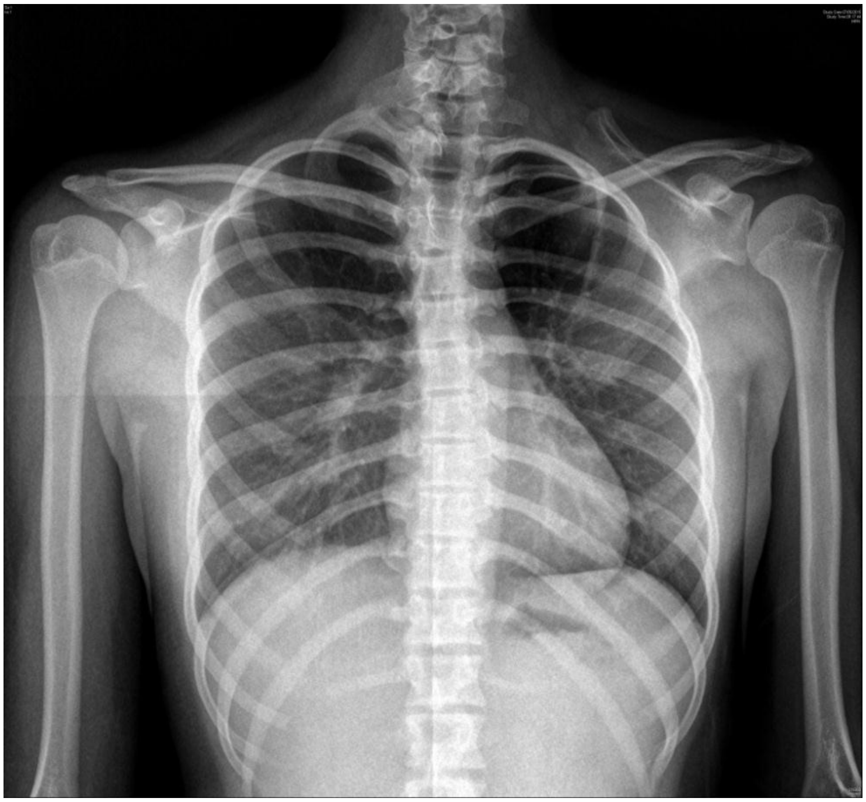


Figure 31.X-ray showing elevated scapula on left side pre-op with Rigualt’s grade-2 of a 8years old child

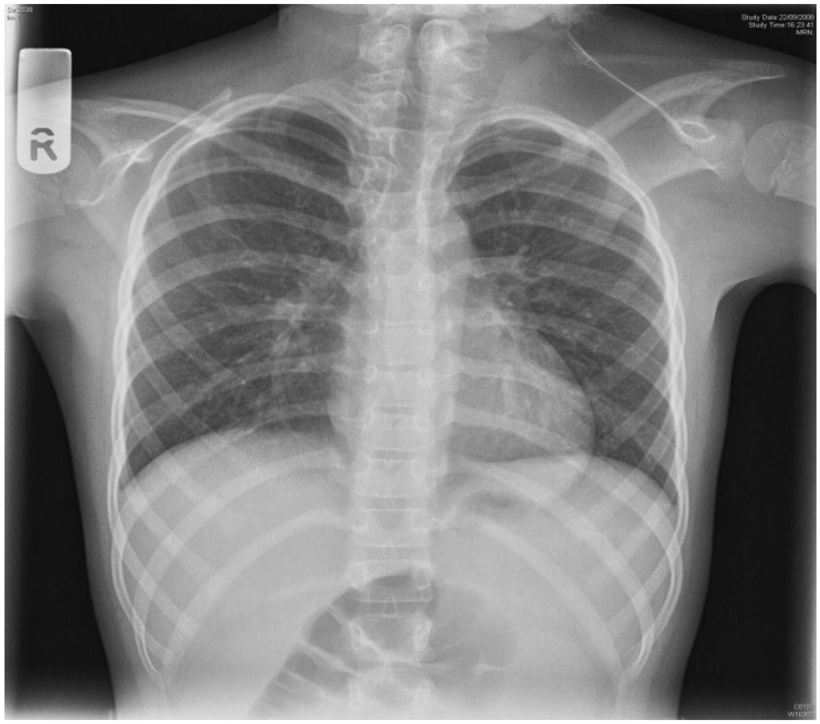


Figure 32 Post-op X-ray showing Rigualt's grade-1 at follow-up following modified Woodward's procedure

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ANNEXURES

PROFORMA

PROFORMA

STUDY TILE: A Retrospective observational study to assess the post operative functional outcomes following Modified Woodward's procedure in patients with Sprengel's deformity.

STUDY NO:

DATE:

NAME: HOSP. NO.:

ADDRESS:

PHONE NO.

DATE OF BIRTH:

AGE: SEX:

SIDE OF OPERATION:

DOMINANT HAND:

AGE AT SURGERY:

PRE OP EVALUATION

	RIGHT	LEFT
ROM ABDUCTION FORWARD FLEXION		

CAVENDISH SCORE		
RIGUALT SCORE		

AGE AT FOLLOW-UP:

RISK FACTORS	PRE-OP	POST-OP
Omovertebral Bone		
Scoliosis		
Hemivertebrae		
Klipel- fiel Syndrome		
Torticolis		

Cavendish Score	Sprengel's deformity
Grade 1 - Very mild.	Shoulder joints are level. Deformity is invisible when patient is dressed.
Grade 2 - Mild.	Lump in the web of the neck. Deformity is visible when dressed.
Grade 3 - Moderate.	Shoulder elevation 2-5 cm. Deformity is easily visible.
Grade 4 - Severe	Superior angle of the scapula is near the occiput with/without neck webbing or brevicollis

Rigault's Classification	
Grade 1	Under Th-1
Grade 2	between Th1, and C5
Grade 3	above C5.

POST OP:

	RIGHT	LEFT
ROM ABDUCTION FORWARD FLEXION		
CAVENDISH SCORE		
RIGUALT SCORE		

RADIOGRAPHIC ASSESSMENTS OF SCAPULA	Normal Side	PRE-OP POST-OP
SUPERIOR DISPLACEMENT RATIO		
HEIGHT TO WIDTH RATIO		
SCAPULAR LOWERING		

QUESTIONNARE FORMAT:

PODCI SCORE:

During the last week was it easy or hard for your child to:

QUESTIONNAIRE	RIGHT					LEFT				
	Easy	Little hard	Vey Hard	Can't do all	Too young for this activity	Easy	Little hard	Vey Hard	Can't do all	Too young for this activity
	1	2	3	4	5	1	2	3	4	5
1.Lift heavy books?										
2. Pour a half gallon of milk?										
3.Open a jar that has been opened before?										
4.Use a fork / spoon										
5.Comb his/her hair?										
6.Button buttons?										
7.Write with a pencil?										
8.Turn door knobs?										

SCORES		RESULT
Raw Score:	Sum of items Q1, Q2, Q3, Q4, Q5, Q6, Q8, Q32	
Mean of Items:	(sum of items Q1, Q2, Q3, Q4, Q5, Q6, Q8, Q32/ (number of non-missing items)	
Standardized Score:	[(4 - mean of items) / 3] * 100	
Normative Score:	10 * [(Standardized score - General population score) / General population standard deviation] + 50.	

Score:

SIMPLE SHOULDER TEST:

CURRENT PROBLEM	RIGHT YES/NO	LEFT YES/NO
1. Is your shoulder comfortable with your arm at rest by your side?		
2. Does your shoulder allow you to sleep comfortably?		
3. Can you reach the small of your back to tuck in your shirt with your hand?		
4. Can you place your hand behind your head with the elbow straight out to the side?		
5. Can you place a coin on a shelf at the level of your shoulder without bending your elbow		

6. Can you lift one pound (a full pint container) to the level of your shoulder without bending your elbow		
7. Can you lift eight pounds (a full gallon container) to the level of your shoulder without bending your elbow?		
8. Can you carry twenty pounds at your side with the affected extremity?		
9. Do you think you can toss a softball under-hand twenty yards with the affected extremity		
10. Do you think you can toss a softball over-hand twenty yards with the affected extremity		
11. Can you wash the back of your opposite shoulder with the affected extremity?		
12. Would your shoulder allow you to work full-time at your regular job?		

SCORING:

PAIN - If you have pain in your shoulder, how bad is it?

a) Present all the time and unbearable; needing strong medications frequently
b) Present always and bearable; strong medications occasionally.
c) No pain or little at rest, but present during light activities and needing mild medications frequently
d) Present during heavy or particular activities only; needing mild medications occasionally.
e) Slight pain occasionally.
f) No pain

FUNCTION - Do you have any restrictions of your shoulder function?

a) Unable to use limb because of this shoulder.
b) Only light activities possible because of this shoulder.
c) Able to do light work/housework or most activities of daily living
d) Most work/housework, shopping and driving possible; able to do hair and dress, undress including reaching behind your back high enough to do up a bra.
e) Slight restrictions only; able to work above shoulder level
f) Normal activities

ACTIVE FORWARD FLEXION - How high up can you left your arm forwards?

a) Greater than 150 degrees
b) 120-150 degrees
c) 90-120 degrees
d) 45-90 degrees
e) 30-45 degrees
f) less than 30 degrees

STRENGTH OR FORWARD FLEXION **-** How strong is your arm?

a) Normal Strength
b) Good strength – a bit weaker
c) fair strength – moderately weaker
d) poor strength – much weaker
e) muscle contraction only
f) nothing

SATISFACTION OF THE PATIENT - Since the operation.

a) Satisfied and better

b) Dissatisfied and worse

Score:

sno	ca_pre	ca_post	rig_pre	rig_post	sup_pre	sup_post	n	ratio_pre	ratio_post	scap_lower	sex
1	3	1	3	2	0.32	0.16	0.93	0.82	0.93	2.3	F
2	3	2	2	1	0.49	0.3	0.9	0.87	0.89	3.4	F
3	4	2	3	2	0.31	0.22	0.98	0.95	0.97	1.8	F
4	4	1	3	1	0.55	0.42	0.85	0.82	0.84	2.8	F
5	3	1	2	1	0.4	0.21	1.01	0.82	0.97	1.8	M
6	3	1	2	1	0.43	0.35	0.92	0.89	0.93	1.5	F
7	3	2	2	1	0.44	0.27	0.93	0.9	0.94	1.4	F
8	4	2	3	1	0.54	0.31	1.06	0.91	0.98	2.1	M
9	4	1	3	1	0.33	0.27	1.03	0.98	1.02	3.8	F
10	4	2	3	2	0.41	0.32	0.97	0.92	0.97	1.6	F
11	4	2	3	1	0.26	0.31	0.9	0.75	0.82	0.9	M
12	3	1	2	1	0.35	0.24	0.96	0.91	0.96	1.4	F
13	3	1	2	1	0.38	0.32	0.97	0.91	0.98	2.1	F
14	3	1	2	1				1.2	1.1	1.3	F

sno	age_sur	Age atfollowup	side_of surgery	anomalies	Omovertebral bar	abduction_pre	abduc_post	ff_pre	ff_post	podci
1	7	10	LEFT		Yes	120	160	130	160	62
2	12	8	LEFT		Yes	110	150	130	160	100
3	4	8	LEFT	Webbed Neck Scoliosis	Yes	120	140	130	160	84
4	3	9	RIGHT	Scoliosis	Yes	100	140	110	150	88
5	5	6	LEFT		Yes	110	150	130	160	63
6	7	10	LEFT		No	130	160	140	170	84
7	5	12	RIGHT		Yes	100	150	100	160	92
8	3	11	RIGHT	Scoliosis	Yes	90	140	100	160	92
9	8	15	LEFT	Scoliosis	Yes	130	160	140	170	100
10	7	15	LEFT	KlippelFeil syndrome	No	120	140	110	140	88
11	5	7	RIGHT	Scoliosis, Congental Vertebral Anomalies ® Anterior Chest Wall Deficiency	Yes	70	100	80	120	84
12	3	5	LEFT		No	90	130	80	140	58
13	6	7	LEFT		No	90	130	100	150	84
14	5	13	RIGHT		Yes	120	160	130	160	92